Anatomical Segmentectomy with a Hybrid VATS Approach in a Patient with Intralobar Pulmonary Sequestration after Severe Pneumonia: A Case Report

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Abstract

Anatomical segmentectomy is an advantageous procedure because it spares healthy lung that has potential to show compensatory growth after lung resection and decreases the risk of air leak and residual resection, which becomes a problem in wedge resection. However, anatomical segmentectomy has not become a common procedure in pediatrics because it requires more complicated procedure than lobectomy or wedge resection, especially in patients with a history of pulmonary infection. In this case report, anatomical basal segmentectomy was safely performed with magnified vision by a hybrid video-assisted thoracic surgery (VATS) approach in a 6-year-old girl with intralobar pulmonary sequestration after severe pneumonia. The result suggests that the indications for hybrid VATS segmentectomy can expand further to include segmental lesions in children.

Keywords

- pulmonary sequestration
- anatomical segmentectomy
- video-assisted thoracic surgery

New Insights and the Importance for the Pediatric Surgeon

Anatomical basal segmentectomy using a hybrid VATS approach was successfully performed in a child with intrapulmonary sequestration following a severe pneumonia. Hybrid VATS approach enables safety segmentectomy in children with magnified vision of thoracoscopy.

Introduction

Anatomical segmentectomy is an increasingly attractive alternative to lobectomy in selected adult patients.[1,2] In children, however, the procedure is less common because of its technical complexity. Segmentectomy requires dividing the segmental bronchus and pulmonary vessels at the hilum. A history of pulmonary infection further increases the risk of complications.[3]

Here, we present the case of a successful anatomical basal segmentectomy in a child with intralobar pulmonary sequestration (IPS) and recent pneumonia, using the invasive hybrid video-assisted thoracic surgery (VATS) approach.[4]

Case Report

A 6-year-old girl with a history of congenital cystic lung disease was referred to our hospital with persistent cough and fever

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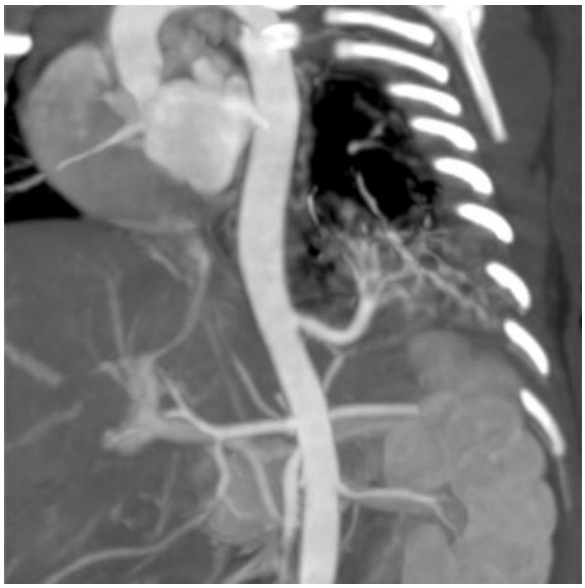

Fig. 1 Postnatal-enhanced computed tomography showing an anomalous arterial supply from the descending aorta to the left lower lobe.

refractory to oral antibiotic therapy. She was diagnosed antenatally with IPS in the left lung with anomalous arterial supply to the left basal segment (►**Fig. 1**). She had no other symptoms prior to admission. Chest computed tomography (CT) showed diffuse consolidation of the left lower lobe. A diagnosis of bacterial pneumonia was made, and intravenous ceftriaxone was initiated. She was discharged after 5 days of treatment.

High-resolution CT showed a cystic lesion limited to the basal segment of the left lower lobe. The apical segment was hypertrophic, suggesting compensatory growth, with normal arterial perfusion, normal venous drainage, and a normal bronchial connection (►**Fig. 2**). A diagnosis of IPS was confirmed, and elective basal segmentectomy with a hybrid VATS approach was planned.

Surgery was performed under general anesthesia with selected left bronchial occlusion, using a Fogarty catheter. The patient was placed in the right decubitus position. A 10-mm thoracoscopic trocar was inserted through the ninth intercostal space in the left mid-axillary line. Thoracotomy was performed through the fifth intercostal space with a 5-cm posterolateral incision. Inflammatory adhesions between the left lung and the chest wall were dissected under thoracoscopic vision. An aberrant artery running through the pulmonary ligament was detected. This was divided by silk thread ligation, clipping, and division using a bipolar system (the LigaSure vessel sealing system (Valleylab/Tyco Healthcare) (►**Fig. 3a**). The common basal vein was identified and divided posteriorly.

Following division of the major fissure, the basal arteries were cut individually (►**Fig. 3b**). Following confirmation of aeration in the apical segment, the basal bronchus was clamped and transected with an endoscopic linear stapler. The lung parenchyma was divided with the stapler on the inflation–deflation line between the affected basal segment and the healthy apical segment (►**Fig. 4**). The specimen was retrieved through the thoracotomy without extension of the wound. The size of the specimen was 10 × 9 cm. The operative time was 217 minutes, and the estimated blood loss was 56 mL.

Histopathological examination revealed a cystically dilated bronchus with inflammation of the surrounding parenchyma. The margin of the specimen did not include cystic lesion, confirming complete resection of the lesion. The wall of the aberrant artery was composed of elastic fibers. These findings were compatible with a diagnosis of IPS.

The patient recovered without complications and was discharged home on postoperative day 5. A chest radiograph 4 months postprocedure demonstrated satisfactory expansion of residual lung (►**Fig. 5**). No respiratory impairment was reported throughout the 1-year follow-up period.

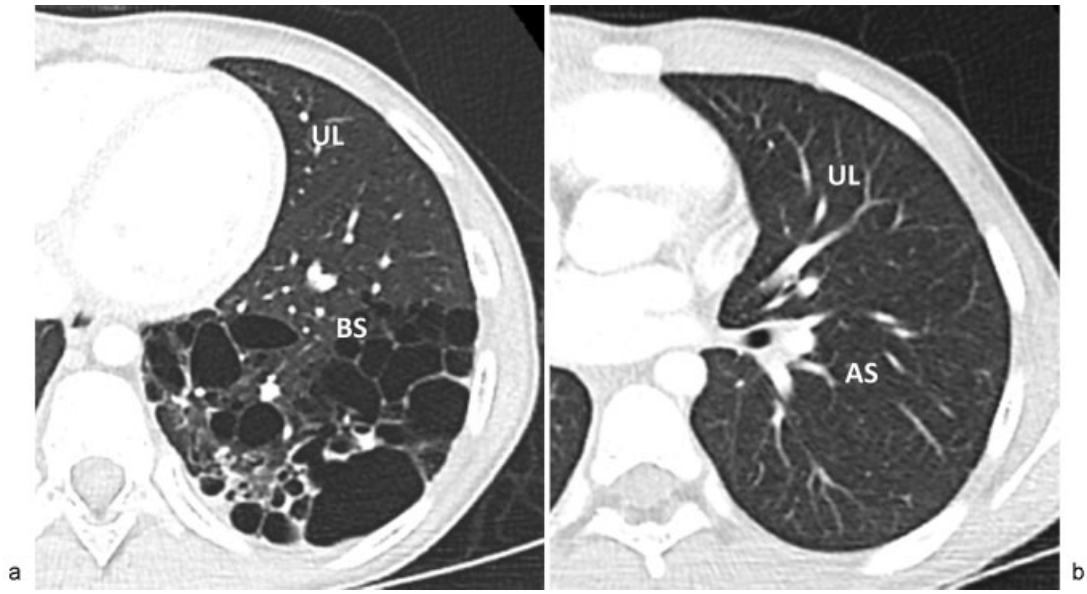

Fig. 2 Preoperative high-resolution computed tomography showing the affected BS with a normal AS. (**a**) UL, BS. (**b**) UL, AS. AS, apical segment; BS, basal segment; UL, upper lobe.

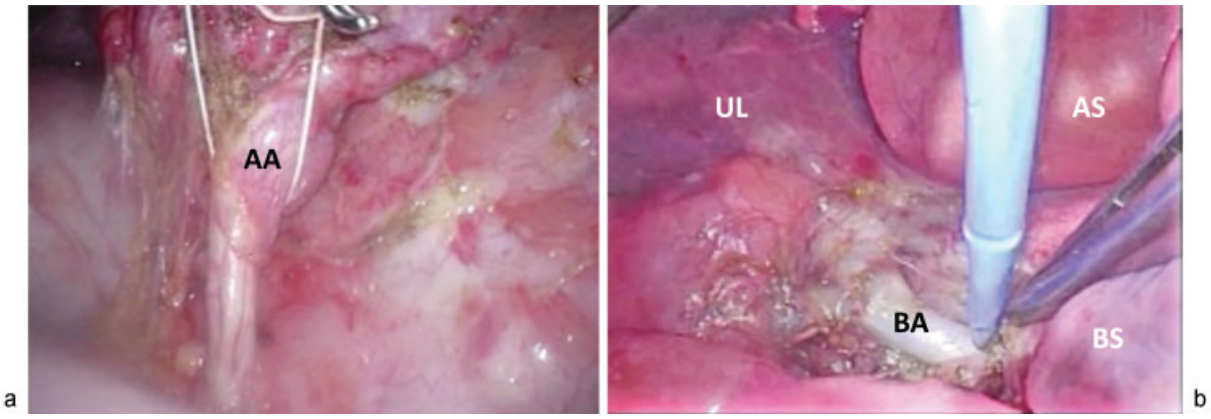

Fig. 3 Intraoperative view. (**a**) AA and (**b**) UL, AS, BS, BA. AA, aberrant artery; AS, apical segment; BA, basal artery; BS, basal segment; UL, upper lobe.

Discussion

Pulmonary sequestration is a congenital lung malformation defined as nonfunctional lung tissue without a bronchial connection to normal lung.[5] In most cases, the abnormal lung tissue is supplied through an anomalous artery, usually arising from the descending aorta. This rare condition accounts for ~1% of all congenital lung malformations. There are two subtypes, characterized according to their pleural covering: IPS, which is surrounded by the neighboring lung's pleura and extralobar pulmonary sequestration, which has its own pleura.

Definitive treatment of IPS is surgical resection to eliminate the possibility of recurrent respiratory infection, malignant transformation, and sudden hemoptysis caused by a ruptured anomalous artery.[6]

Lobectomy has been the most common surgical approach to IPS, as it is technically simple and affords reliable confirmation of complete resection. As recent advances in radiology have enabled surgeons to understand the detailed pulmonary anatomy preoperatively, lung-sparing surgeries such as segmentectomy and wedge resection are now considered viable options.[3]

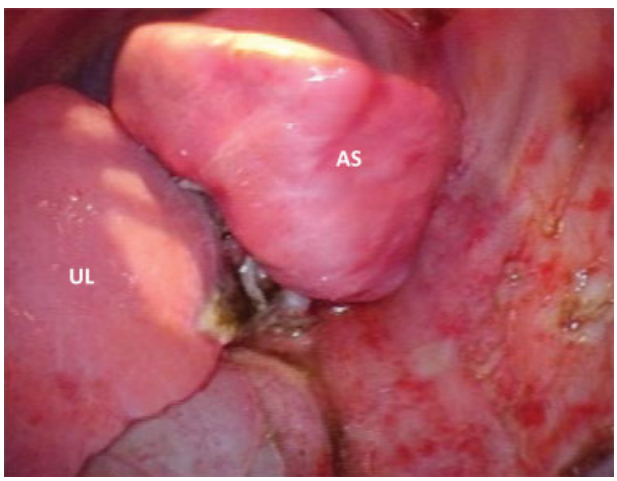

Fig. 4 Thoracoscopic view after basal segmentectomy showing preserved healthy apical segment. AS, apical segment; UL, upper lobe.

Anatomical segmentectomy is preferable to lobectomy as the former spares lung that has potential for compensatory growth following lung resection. One report suggested decreased exercise tolerance of children following lung resection.[7] In older children, we do not expect a great deal compensatory lung growth following lung resection. Nevertheless, sparing healthy lung is valuable for preserving overall respiratory function, particularly if the healthy lung is hypertrophic, as in our case.[8] The preserved lung can prevent thoracic deformity by expanding into the space created by resection.[9] Finally, defined anatomical segmentectomy decreases the risk of residual lesion that often results from atypical resections.[3,10]

Another lung-sparing surgical approach is wedge resection, in which the affected lung is nonanatomically resected at the demarcation line. In wedge resection, sufficient space and margins are required to clamp the parenchyma with a surgical stapler. In the present case, the lesion was too large, and there was not adequate space to staple the parenchyma without risk of injuring hilar structures. The lesion occupied almost the entire basal segment, making it difficult to distinguish diseased tissue from normal parenchyma. We felt wedge resection risked leaving residual lesion. Therefore, we opted for anatomical segmentectomy.

Anatomical segmentectomy in this case presented several challenges. First, the patient had a history of pneumonia that might have caused severe adhesions in the thoracic cavity. Second, unlike other congenital lung malformations, resection of an IPS necessitates division of an anomalous artery. This requires meticulous manipulation deep in the thoracic cavity. To solve these problems, we employed a hybrid VATS approach with satisfactory results.

The efficacy of thoracoscopy in pediatric patients has been well documented over the past 20 years.[11] Thoracoscopic surgery is a safe and effective procedure. The procedure decreases postoperative pain and length of hospital stay.[12] However, the port-access feature limits the direction of view and mobility of surgical instruments. Therefore, thoracoscopic surgery tends to be avoided in complicated cases, such as newborns with comorbid congenital anomalies or respiratory infections.[13,14]

Hybrid VATS is a variation of thoracoscopic surgery in which the procedure is performed using a combination of

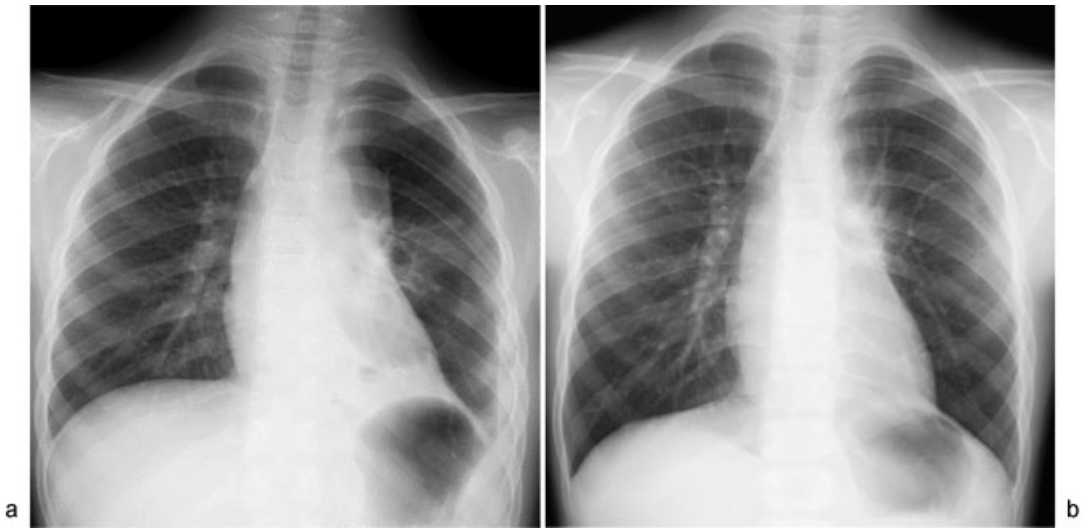

Fig. 5 Postoperative chest radiograph (**a**) 5 days after surgery and (**b**) 4 months after surgery.

direct view through a utility thoracotomy and magnified view via thoracoscopy.

Excision of an IPS entails the risk of tearing the aberrant artery and subsequent life-threatening hemorrhage.[15] In our case, under magnification, the aberrant artery was easily distinguished from surrounding inflammatory tissues, and meticulous division was achieved despite the presence of a severe pneumonia.

There are reports of pediatric lung surgery employing a complete VATS approach without the use of a utility thoracotomy.[16] In our case, a complete VATS approach might have been unsuitable, as the lesion was too large to retrieve via a port site. We believe a 5-cm utility thoracotomy was necessary as well, as we required meticulous dissection and separation of tissues in a small and deep thoracic space.

There is some debate over timing of surgery for congenital pulmonary malformations, especially regarding risk in the presence of recent pneumonia.[17] Rather than performing surgery when our patient was young and small, we adopted a "wait-and-see" approach, and delayed surgery until the infected sequestration became infected and was successfully treated. By this time, the patient was older and larger, both conditions being more favorable for resection. The patient's lung had healed from pneumonia well enough that the anatomical margin was easily distinguishable. This made less-invasive segmentectomy more feasible. Although we found some adhesions between the lung and the parietal pleura, these could be dissected safely under thoracoscopic vision.

Conclusion

We safely performed an anatomical basal segmentectomy with magnified vision using a hybrid VATS approach in a child with IPS following a severe pneumonia. Our results suggest that the indications for hybrid VATS segmentectomy may be expanded further to include segmental lesions in children.

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