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CASE REPORT

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Abnormal arterial ligation alone for patients with anomalous systemic arterial supply to the left basal segment of the lung: three case series

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Abstract

Background Anomalous systemic arterial supply to the left basal segment of the lung is a rare congenital pulmonary vascular malformation, historically classified as a variant of intra-lobar pulmonary sequestration. The standard surgical approach has typically involved ligation of the anomalous artery in combination with lobectomy or segmentectomy.

Case description We present three cases of anomalous systemic arterial supply to the left basal segment of the lung, all successfully treated with thoracoscopic anomalous arterial ligation alone. In one case, indocyanine green was used to assess the blood supply, enhancing procedural safety.

Conclusion Thoracoscopic ligation of the anomalous artery alone may be a safe and lung-preserving surgical option for patients with this rare anomaly. The use of indocyanine green can further improve the safety of the procedure.

Keywords Anomalous systemic arterial supply to the left basal segment of the lung, Intra-lobar pulmonary sequestration, Arterial ligation, Indocyanine green

Background

Anomalous systemic arterial supply to the left basal segment of the lung (ASALLL) is a rare congenital malformation of the pulmonary vasculature, previously classified as type I pulmonary sequestration by Pryce et al. [1]. However, it is now recognized as a distinct entity from intra-lobar pulmonary sequestration (ILS) due to the preservation of normal bronchial and venous anatomy in the affected lung lobe. The treatment approach for ASALLL remains debated, with traditional management often mirroring that for ILS, including anomalous artery ligation and lobectomy or segmentectomy. However, if ASALLL patients are suitable candidates for isolated anomalous arterial ligation, this less invasive approach would help preserve lung function and improve long-term quality of life. In this report, we describe three cases of ASALLL successfully treated with thoracoscopic anomalous arterial ligation, with all patients resuming their preoperative daily activities and achieving favorable postoperative outcomes.

Case description

Case 1

A 32-year-old female with ASALLL presented with a four-month history of hemoptysis. Preoperative chest computed tomography (CT) revealed an anomalous

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arterial branch arising from the thoracic aorta (Fig. 1A, B). Unlike patients with ILS, her left lower lobe exhibited no signs of pneumonia or consolidation. In the course of surgery, after assessing lung viability, we opted to preserve the left lower lobe. The patient has been followed for eight years, during which annual chest CT scans showed no evidence of atrophy in the lung lobe (Fig. 1C, D).

Case 2

A second 32-year-old female presented with a one-month history of hemoptysis. Preoperative enhanced chest CT and three-dimensional reconstruction confirmed ASALLL without any architectural distortion (Fig. 2A, B). The patient and her family requested definitive treatment to control her symptoms while preserving lung lobe as possible. Consequently, we opted for thoracoscopic arterial ligation, with the intraoperative decision made regarding the preservation of the diseased left lower lobe. A large anomalous artery supplying the left lower lobe was identified and ligated, with no significant signs of ischemia observed in the lung tissue, thus allowing preservation of the lobe.

However, on postoperative seventh day, the patient developed a fever, and chest CT revealed pneumonia in the left lower lobe. Symptoms improved with conservative antibiotic therapy. The patient has been followed for ten months, during which there has been no recurrence of pneumonia, hemoptysis, or other pulmonary symptoms. Chest CT at eight months postoperatively showed mild atrophy of the left lower lobe but no evidence of parenchymal inflammation or consolidation (Fig. 2C). The patient has since resumed normal work and daily activities.

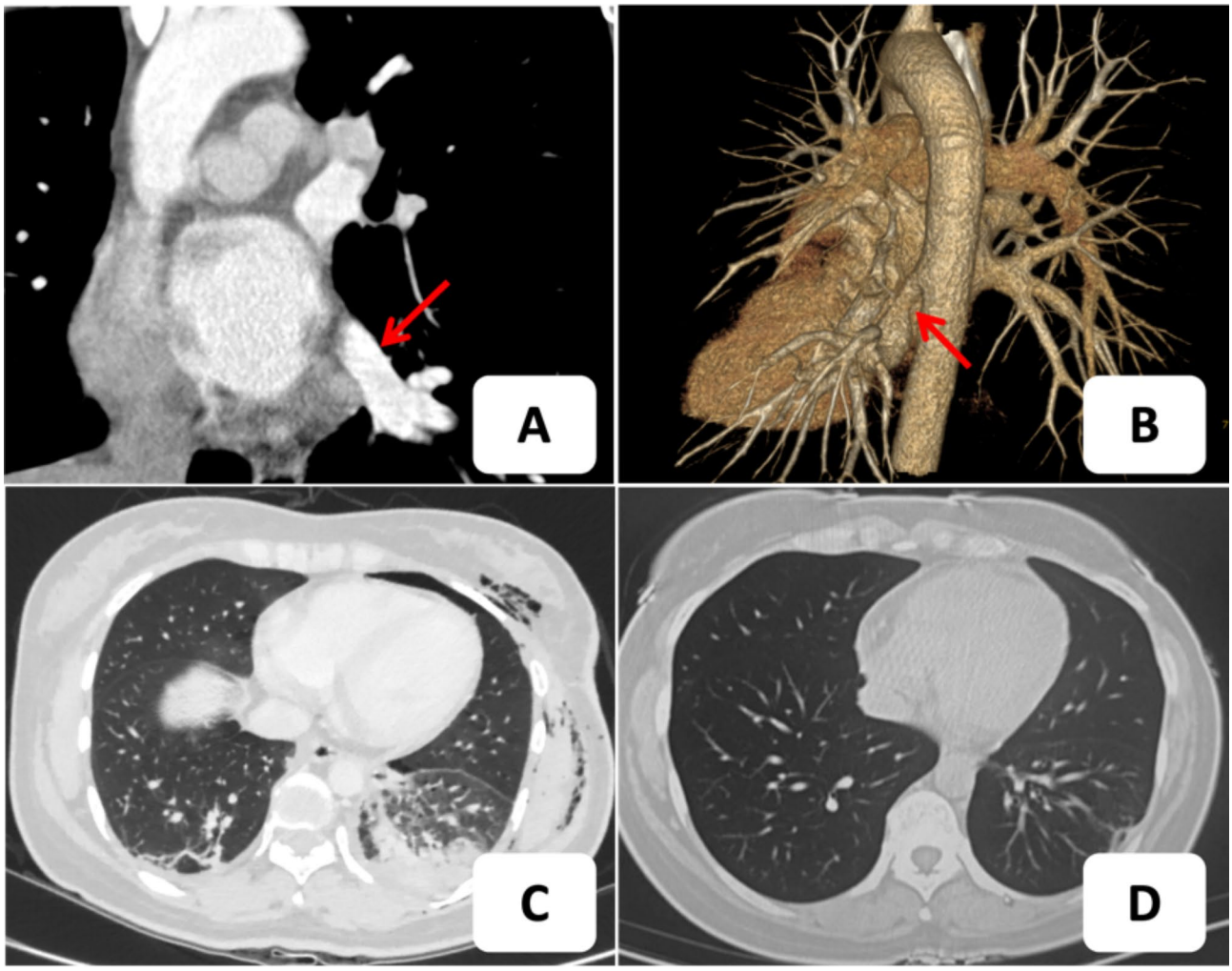

Fig. 1 Chest Radiographic Images of Case 1 **A**,**B**: Preoperative contrast-enhanced chest CT and three-dimensional CT reconstruction, revealing an abnormal artery originating from the thoracic aorta. **C**, **D**: Postoperative follow-up imaging of Case 1. Chest CT at three months post-surgery shows exudative changes in the left lower lobe, while chest CT at eight-year post-surgery shows no structural changes in the left lower lobe, with only mild atrophy observed

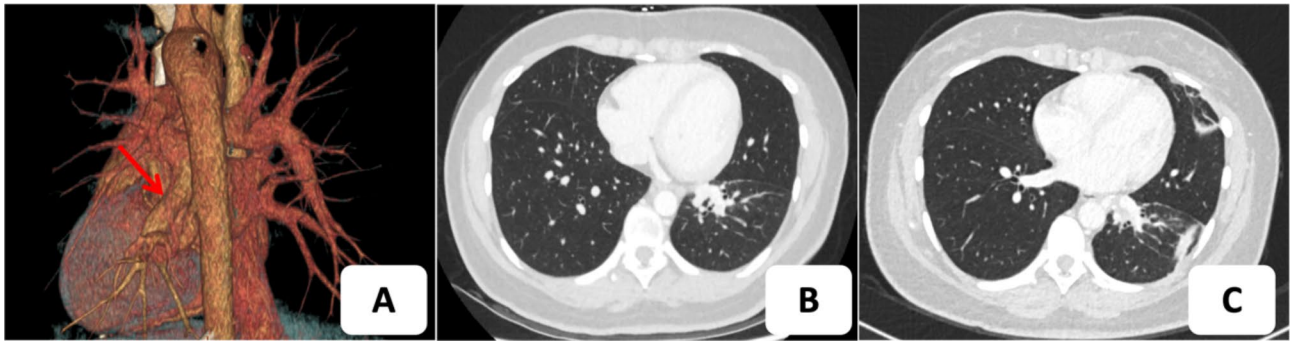

Fig. 2 Chest Radiographic Images of Case **2 A**: Preoperative three-dimensional CT reconstruction reveals an abnormal artery originating from the thoracic aorta. **B**: Preoperative contrast-enhanced chest CT shows not only the abnormal artery but also thickened vascular shadows. **C**: A contrast-enhanced chest CT scan at eight months post-surgery reveals mild shrinkage of the left lower lobe and linear bands within the lobe, without signs of pneumonia or consolidation

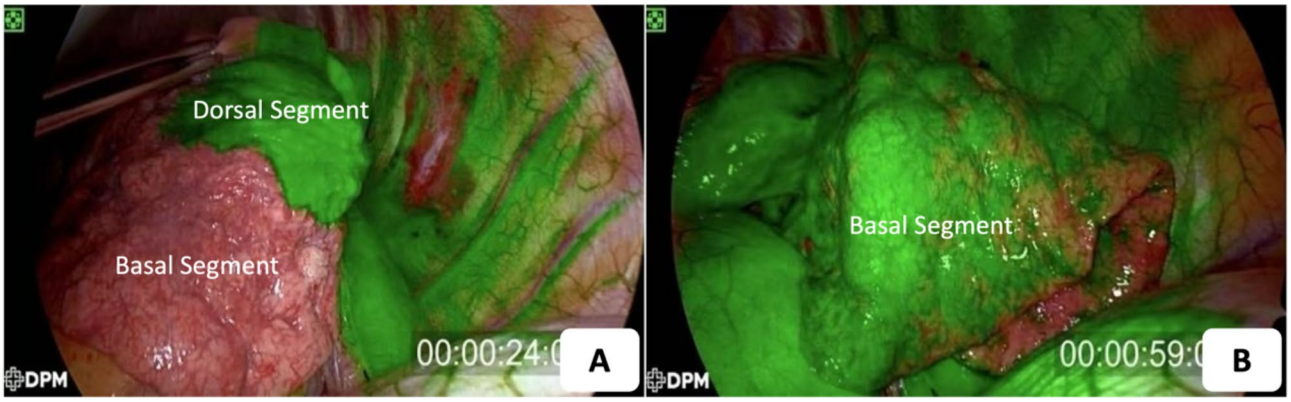

Fig. 3 Intraoperative fluorescence imaging in Case 3 **A**: In the first thirty seconds after intravenous injection of ICG, fluorescence is visible in the dorsal segment of the left lower lobe, but not in the basal segment of the left lower lobe. **B**: Approximately one minute later, a rapid and uniform fluorescence was observed in the basal segment

Case 3

A 28-year-old female with ASALLL presented with intermittent hemoptysis for approximately one year. The patient and her family opted for thoracoscopic anomalous artery ligation. Following transection of the anomalous artery using a linear stapler, 20 mg of indocyanine green (ICG) was administered intravenously, and fluorescence microscopy was employed to assess the fluorescence imaging pattern. This approach was intended to confirm the presence of collateral blood supply to the left lower lobe, which would support the decision to preserve the affected lobe. Approximately thirty seconds after ICG injection, fluorescence was observed in both the normal left upper lobe and dorsal segment of the left lower lobe, followed by rapid and uniform fluorescence enhancement in the basal segment of the left lower lobe (Fig. 3). This finding indicated collateral blood supply to the left lower lobe, prompting the decision to preserve it. The patient has been followed for six months without recurrence of symptoms or any significant discomfort.

Discussion

ASALLL is a distinct disease characterized by the isolated left lower lobe receiving blood supply from an anomalous systemic artery while maintaining normal connections to the bronchial and venous systems. Preoperative contrast-enhanced chest CT scans can reveal aberrant artery originating from the systemic circulation, often accompanied by thickening of the lung markings in the left lower lobe. This differs from patients with ILS, whose affected lung lobe frequently undergoes recurrent infections due to structural abnormalities, such as bronchiectasis [2]. Lung ventilation-perfusion (V/Q) imaging often shows no abnormal areas in ASALLL patients, distinguishing them from other forms of pulmonary sequestration. The relatively normal parenchyma in ASALLL patients is a key factor in deciding whether to preserve lung tissue. Additionally, determining whether the affected lobe has an alternative blood supply is crucial for surgical decision-making regarding lung preservation.

The preferred surgical approach for ASALLL remains a combination of anomalous arterial ligation with lobectomy or segmentectomy. Although this approach

effectively addresses symptoms in the short term, there are still concerns about the long-term impact of excessive resection of lung tissue on quality of life, especially in young patients.

Endovascular embolization, using devices such as the Amplatzer, has also been reported for managing ASALLL. However, after consulting with our interventional vascular surgery team, we still chose surgical ligation for these patients. Our primary concern was the feasibility of complete embolization, given that the anomalous artery originated from the high-pressure descending aorta or thoracic aorta. Furthermore, the risk of device displacement, potentially leading to lung infarction and complicating subsequent surgical procedures. Several studies have highlighted severe postoperative complications following endovascular embolization [3–5]. These factors ultimately led us to opt for surgical ligation.

The literature on thoracoscopic anomalous arterial ligation for ASALLL is limited [6–8], with few systematic reviews addressing postoperative recovery and long-term outcomes. Thoracoscopic arterial ligation offers several benefits, including preservation of pulmonary function, reduced surgical trauma, and fewer severe postoperative complications. If collateral circulation is established or additional blood supply from the anomalous artery is available, the preserved lung lobe may regain partial gas-exchange function.

In this report, we present three cases of ASALLL treated with thoracoscopic anomalous arterial ligation. Preoperative pulmonary function test (PFT) for Case 2 and Case 3 revealed FEV1/FVC ratio of 77.93% and 82.98%, respectively, and DLCO-SB value of 6.42 mmol/min/kPa and 7.16 mmol/min/kPa, indicating normal ventilatory function but slightly reduced diffusing capacity. During the surgery for Case 3, we utilized near-infrared fluorescence imaging to assess blood supply to the left lower lobe, thereby enhancing procedural safety. This approach enabled real-time evaluation of blood perfusion and supported the decision to preserve lung tissue. Postoperatively, all three patients resumed normal daily activities. However, a limitation of our follow-up is the lack of routine postoperative PFT, which prevented a quantitative assessment of lung function recovery.

Case 1 represents the ASALLL patient with the longest follow-up. Eight years post-surgery, the left lower lobe showed minimal atrophy, and the patient's exercise tolerance remained unchanged from pre-disease levels. This suggests that the diseased lung regained some functional blood-air exchange.

Case 2, who experienced transient pneumonia, showed mild lung atrophy but has since returned to normal activity levels. No signs of pneumonia or consolidation were noted in the preserved lobe. We consider the pneumonia was due to transient ischemia caused by the loss of primary blood supply to the left lower lobe, rather than infection related to structural abnormalities such as bronchiectasis. This case underscores the importance of intraoperative assessment of blood supply and careful postoperative care to prevent pneumonia following lung-preserving surgery in ASALLL patients.

Case 3 underwent lung-preserving surgery after identifying an alternative blood supply to the affected lobe during the procedure. Six months postoperatively, the patient remains asymptomatic. During surgery, intravenous ICG was used to assess blood supply, a novel application not previously documented in the literature. This technique, adapted from ICG-guided segmental lung resection [9], enabled the identification of collateral blood supply via other arteries such as bronchial arteries. After ligating the anomalous artery, a rapid and uniform fluorescence pattern was observed, indicating additional blood supply to the affected lung lobe, as this fluorescence pattern was distinct from the slower fluorescence diffusion through lung tissue. This novel application of ICG provides an efficient and convenient means to assess blood perfusion and may have broader applications in ASALLL surgery.

Conclusion

We present three ASALLL cases treated with thoracoscopic anomalous arterial ligation, a safe approach for patients with mild pulmonary architectural abnormalities, if determined by preoperative evaluation. It may also be applicable to other forms of ILS, provided the lesions are similarly mild. However, further experience is needed to refine and quantify the criteria for selecting suitable candidates for this surgical approach.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s13019-024-03280-y.

Supplementary Material 1

Author contributions

Huiming Han carried out the manuscript and collected references. Hao Li coordinated all authors. Sida Cheng, Hui Zhao and Jianfeng Li underwent this operation, and Fan Yang helped for clinical support with them. Huiming Han and Sida Cheng took pictures of the image and operation. Sida Cheng and Hao Li helped to draft the manuscript. All authors read and approved the final manuscript.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Consent for publication
Informed consent to publish this case report was provided by the patient.

Dual publication
The results/data/figures in this manuscript have not been published elsewhere, nor are they under consideration (from you or one of your Contributing Authors) by another publisher.

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