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CASE REPORT

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Coil embolization of anomalous systemic artery to the left lower lobe in an asymptomatic adult: a case report

Meng Sun[1†], Le Fang[2†], Fangruyue Wang[3] and Tianwei Wang[1*]

Abstract

Background Anomalous systemic artery to the left lower lobe (ASALLL) is a rare congenital anomaly. The primary symptoms include hemoptysis and lung infection, though some patients may remain asymptomatic. Currently, there is no consensus on the indications for treatment or the optimal choice of therapy for this condition. This case presents a case of an asymptomatic adult who underwent coil embolization.

Case presentation A 48-year-old male was admitted to our hospital due to the discovery of a space-occupying lesion in the left hilum. The contrast-enhanced pulmonary computed tomography scan was used to diagnose the patient with an anomalous systemic artery to the left lower lobe. We performed coil embolization on this patient, who underwent a follow-up computed tomography angiography of the pulmonary and bronchial arteries one year later. Result demonstrated complete embolization of the abnormal systemic arteries and a slight reduction in the volume of the left lower lobe.

Conclusion Coil embolization is a safe and minimally invasive procedure for adult patients who have an anomalous systemic artery to the left lower lobe.

Keywords Pulmonary vascular malformation, Coil embolization, Lung, Asymptomatic, Rare case

Introduction

Anomalous systemic artery to the left lower lobe (ASALLL) is a rare congenital anomaly. The main characteristic of this condition is an anomalous artery arising from the descending aorta and providing blood supply to the lower lobe of the left lung. The bronchial segments involved undergo normal development, while the corresponding left lower pulmonary artery may be absent or significantly thinner than usual [1]. The etiology of the disease remains uncertain, however, it is likely that the condition is attributable to aberrant degeneration of the primitive aorta, which supplies the lung bud during embryonic development [2]. A review of the literature indicates that ASALLL is more prevalent in Asian populations, with a higher incidence observed in males compared to females [3]. The majority of patients may present with a range of symptoms, with hemoptysis being the most prevalent, followed by chest pain, recurrent infections, congestive heart failure, and other related issues [4]. The main treatment methods for this condition are surgical intervention and interventional embolization [5]. Some patients may remain asymptomatic. Currently, there is no consensus on the indications for treatment or the optimal choice of therapy for this condition. In this

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case, we present an asymptomatic adult male patient in whom an abnormal shadow was detected in the left hilar region during a routine physical examination. Further imaging revealed an anomalous vascular supply to the lower lobe of the left lung originating from the thoracic aorta, along with normal development of the left lower pulmonary artery. Given the potential for hemodynamic changes resulting from this pulmonary vascular malformation to eventually result in pulmonary hypertension, hemoptysis, and heart failure [6], we elected to conduct coil embolization on this patient. No complications were observed following the procedure, and the patient thoroughly recovered. In this case report, we conducted a comprehensive literature review to investigate whether prompt intervention should be undertaken in asymptomatic patients.

Case description

The patient, a 48-year-old male, was admitted following the detection of an abnormal opacity in the left hilar region of the lung during a routine physical examination, despite exhibiting no discomfort. He had no history of lung disease but had long-standing diabetes mellitus, currently managed with insulin therapy, achieving stable glycemic control. Physical Examination: Breath sounds were clear in both lungs, with no crackles or rhonchi observed. Echocardiography: No abnormal flow signals were detected in the tricuspid and pulmonary valves. The internal diameters of the aorta and pulmonary arteries, as well as the dimensions of the left atrium, left ventricle, right atrium, and right ventricle, were all within normal ranges. Complete blood count: Hemoglobin was 167.0 g/L (110.0–150.0 g/L); platelet count was 0.280% (0.108–0.272%); red blood cell count was 5.63×10^6/μL (3.50–5.50×10^6/μL). Tumor marker: Neuron-specific enolase (NSE) was 21.88 ng/mL (<25.00 ng/mL). Contrast-Enhanced Pulmonary computed tomography (CT) scan (Fig. 1): Scattered patchy opacities were observed in the lower lobe of the left lung, indicative of pulmonary congestion, and an abnormal arterial blood supply was noted in the lower lobe of the left lung, originating from the left anterior wall of the thoracic descending aorta at the level of the 8th thoracic vertebra, with a diameter of approximately 10.5 mm; the left lower pulmonary artery

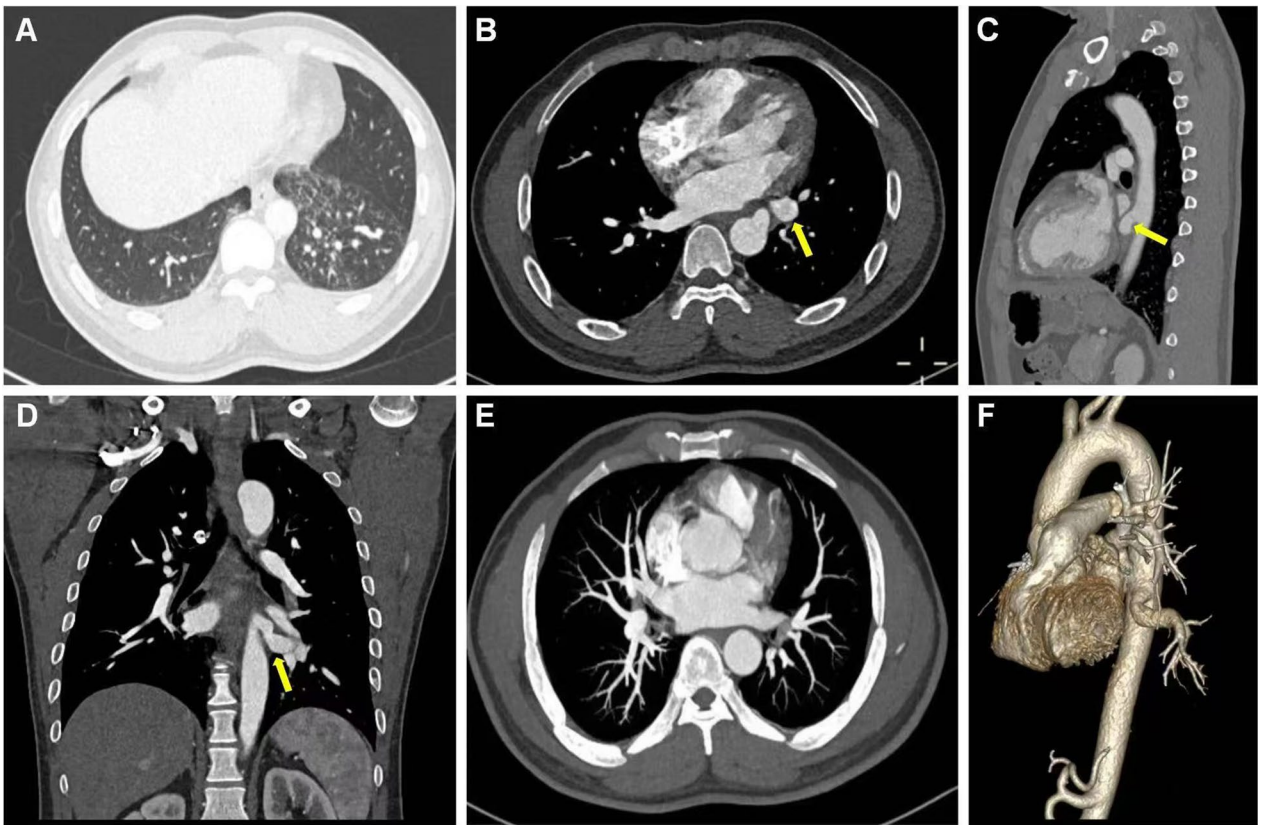

Fig. 1 Preoperative contrast-enhanced pulmonary CT scan: **A** Lung window reveals multiple patchy opacities in the lower lobe of the left lung. **B** Mediastinal window reveals an anomalous artery originating from the thoracic aorta. **C**, **D** MPR reconstruction reveals the anomalous artery accompanied by the left lower pulmonary vein. **E**, **F** MIP (**E**) and VR (**F**) reconstructions demonstrate the anomalous artery originating from the thoracic aorta, with normal development of the left lower pulmonary artery

was normal. Based on the imaging findings from the patient's enhanced CT lung scan, a diagnosis of ASALLL was considered. This condition primarily required differentiation from pulmonary sequestration but also from conditions such as congenital pulmonary arteriovenous fistula, partial anomalous pulmonary venous return, and tracheal Dieulafoy disease.

Following a multidisciplinary consultation involving the departments of thoracic surgery, cardiac surgery, and vascular surgery, the expert panel recommended surgical intervention or coil embolization of the abnormal artery, along with stenting of the descending aorta. Ultimately, we decided to perform the coil embolization on this patient only. Digital subtraction angiography (DSA) was initially performed. A 5-French sheath (Radifocus, Asahi Intecc Co., Ltd., Aichi, Japan), a 2.6-French microcatheter (Stride, Asahi Intecc Co., Ltd., Aichi, Japan), and a 0.018-inch microwire (Stream STM180-18S, Asahi Intecc Co., Ltd., Aichi, Japan) were used. The procedure confirmed a tortuous arterial supply to the left lower lung originating from the descending aorta (Fig. 2A). Subsequently, seven 3D Axium coils (EV3, Medtronic) were implanted, comprising three 16 mm × 40 cm, two 20 mm × 50 cm, and two 18 mm × 40 cm coils. Postoperative combined pulmonary artery and aorta computed tomographic angiography (CTA) (Fig. 3) showed an anomalous artery with striated and patchy low-density filling defects accompanied by high-density coils. No abnormalities were detected in the bilateral pulmonary arteries. One year later, the patient underwent a follow-up computed CTA of the pulmonary and bronchial arteries. The main trunk and distal branches of the abnormal systemic arteries showed no enhancement, confirming complete embolization (Fig. 4A). Upon adjustment to the lung window, a slight reduction in the volume of the left lower lobe was noted (Fig. 4B), with no other significant abnormalities.

Discussion

ASALLL was first classified by Pryce in 1946 as type I intralobar sequestration [7]. Our review of the relevant English literature indicates that approximately 87 cases have been reported to date. In 1974, Sade et al. [8] consolidated several distinct anatomical abnormalities based on Pryce's research, thereby establishing the concept of a disease spectrum that encompasses various combinations of abnormal bronchial connections, aberrant arterial supply, and venous drainage. Clements and Warner [9] refined this classification system in 1987 and introduced the Pulmonary Malinosculation Spectrum, which encompasses congenital abnormal connections involving one or more of the four pulmonary components: airways, arterial blood supply, venous drainage, and parenchyma. The condition can be classified as a type of "arterial blood supply malformation" within this spectrum. The prevailing view among scholars is that this condition is distinct from classic pulmonary sequestration. This is based on the observation that the bronchial tree and lung parenchyma in the affected lung tissue appear normal and that the bronchi are connected to the normal lung. The condition is more frequently observed on the left side, with

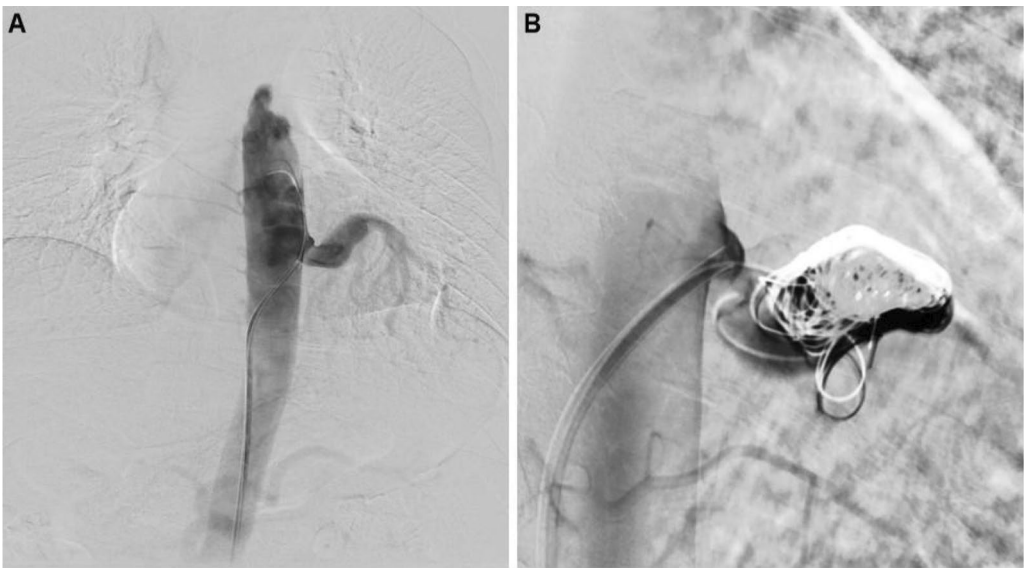

Fig. 2 **A** Intraoperative DSA of the thoracic aorta demonstrates the anomalous systemic artery originating from the thoracic aorta. **B** Intraoperative follow-up imaging after spring coil embolization reveals a spring coil in the proximal lumen of the anomalous artery, with no visualization of the distal portion of the artery

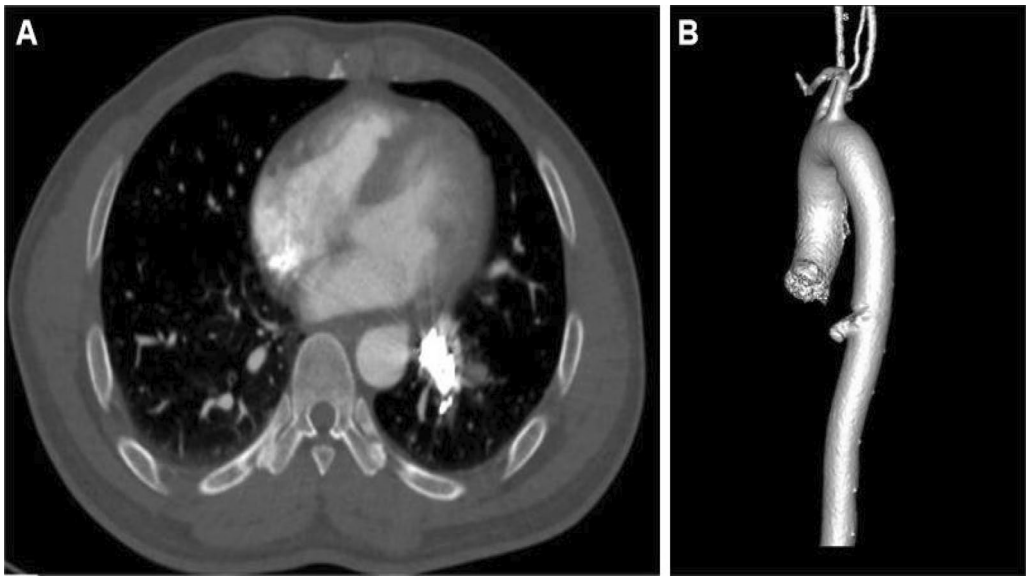

Fig. 3 Combined CTA of the aorta and pulmonary artery on postoperative day 1: **A** Striated and patchy low-density filling defects accompanied by high-density coils in the proximal segment of the anomalous artery. **B** VR reconstruction of the aorta demonstrates that the distal segment of the anomalous artery is not visible, indicating successful embolization

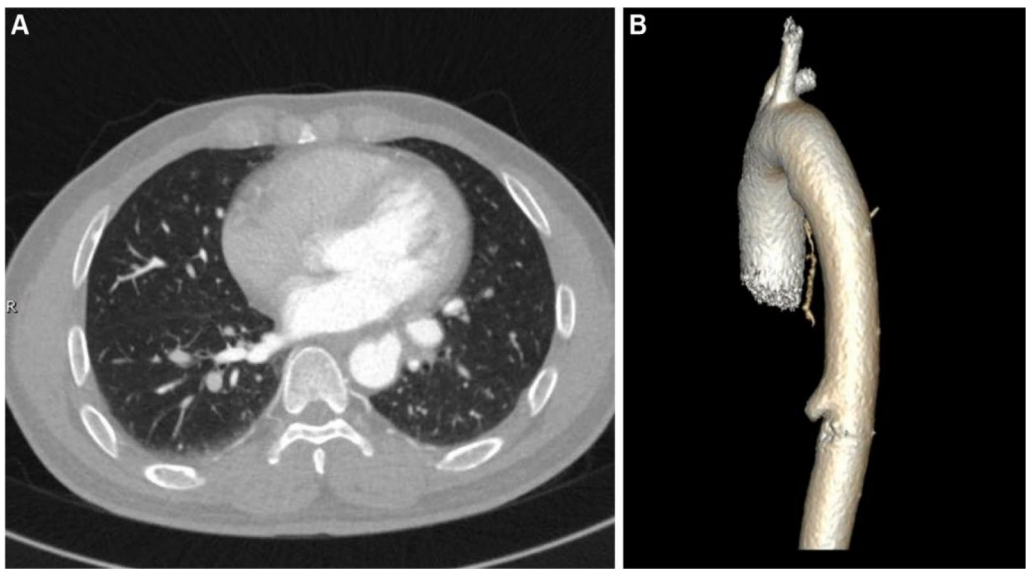

Fig. 4 CTA of the pulmonary and bronchial arteries at 1-year follow-up: **A** The lung window demonstrates a slight reduction in the volume of the left lower lobe; **B** The aortic VR reconstruction reveals no enhancement in the distal portion of the abnormal arteries, confirming complete embolization

only a few cases (4/87) reported in the lower lobe of the right lung. In the majority of ASALLL patients, the blood supply arteries originate from the descending thoracic aorta, with a few cases arising from the coeliac trunk [10]. Typically, the pulmonary artery in the affected lung segment is either absent or thinner than the corresponding pulmonary artery in a healthy lung [11]. When the left lower pulmonary artery is absent, the affected lung segment is solely supplied by an abnormal systemic artery. In this case, the patient's bilateral pulmonary arteries developed normally, and the left lower lobe of the lung was supplied by both the systemic and pulmonary arteries. This is relatively uncommon compared to other reported cases of ASALLL. A review of the literature revealed that

except for cases that did not specifically describe the pulmonary artery, approximately 16% of cases had normally developed pulmonary arteries.

The majority of ASALLL patients present with varying degrees of symptoms. According to our literature review, there were only 19 patients who were asymptomatic (Table 1). The patients' average age was 34 years, with the highest recorded age being 80 years [12, 13]. The smallest patient was only 6 days old [14]. The earliest symptom onset occurred three days after birth [5], while the latest symptom onset occurred at 80 years. The mean age at symptom onset was 31 years. Additionally, the average age of asymptomatic patients was approximately 43 years. The most common manifestation in symptomatic patients is Hemoptysis, occurring in 49% of cases, which is consistent with the findings of Higuchi et al. [15]. High pressure within the supplying arteries is likely to be closely associated with Hemoptysis. Such abnormal hemodynamic change causes an increase in the pressure in the pulmonary vascular bed, which in turn causes rupture of the pulmonary capillaries and their corresponding small veins, leading to hemorrhage, and may be accompanied by the deposition of hemosiderin. Furthermore, patients may develop recurrent respiratory infections, which may be caused by the incomplete obstruction of the bronchi in the afflicted lung tissue by abnormal systemic arteries and pulmonary veins that have thickened due to the increased hemodynamic load. Finally, a persistent left-to-left shunt may lead to congestive heart failure, which is more commonly observed in infants and young children with this condition [16–19]. The severity of the patient's symptoms is primarily influenced by the pressure and diameter of the anomalous arterial vessel. And the average diameter of the anomalous artery is approximately 8 to 10 mm [20]. Some patients may remain asymptomatic due to the small shunt in the anomalous artery [12]. However, aneurysmal dilation may result in a substantial enlargement of the vessels in a small proportion of asymptomatic patients [15, 20]. Approximately 80% of asymptomatic patients exhibit either complete or partial absence of the left lower pulmonary artery (Table 1).

The diagnosis of ASALLL is primarily based on medical imaging. Firstly, chest X-ray can directly observe signs such as nodular or mass shadows that are located posterior to the heart shadow [21]. However, the region posterior to the heart shadow is a blind spot for diagnostics, which could result in misdiagnosis or the overlooking of conditions. Non-contrast CT is a diagnostic method capable of identifying morphological abnormalities in arteries. However, the misdiagnosis of pulmonary space-occupying lesions may result from the presence of an enlarged vascular lumen or aneurysms [22]. In the case of this patient, a space-occupying lesion in the left

Table 1 Review of the literature published in English to date on asymptomatic anomalous systemic artery to the left lower lobe

Year	Author	Age (years)	Diameter (mm)	Pulmonary arterial	Treatment
2020	Utsumi et al. [20]	42	33	Present	Lobectomy and embolization of the remnant artery
2020	Wee et al. [22]	61	11 (with aneurysmal dilatation, 38 cm)	Absent	None
2017	Machida et al. [12]	80	Unknown	Absent	Embolization
2013	Sugihara et al. [6]	67	7	Absent	Embolization
2013	Higuchi et al. [15]	57	25 (with aneurysmal dilatation)	Absent	Lobectomy
2010	Yu et al. [1]	30	19	Absent	Unknown
		28	13	Absent	Unknown
		49	10	Absent	Unknown
		29	20	Absent	Unknown
		32	14	Absent	Unknown
2005	Agarwal et al. [13]	64	10	Unknown	Unknown
2003	Iizasa et al. [41]	20	8	Absent	Lobectomy
2001	Ashizawa et al. [24]	29[#]	Unknown	Absent	Surgery
2000	Ko et al. [23]	47	Unknown	Present	None
		58	Unknown	Absent	None
1999	Yamanaka et al. [2]	30	8	Absent	Anastomosis
1998	Brühlmann et al. [36]	51	11	Present	Embolization
1996	Hirai et al. [42]	30	8	Absent	Anastomosis
1993	Kurosaki et al. [43]	51[#]	Unknown	Absent	Lobectomy/Segmentectomy

[#] Mean

hilar region was observed during an initial non-contrast CT scan. Consequently, the patient was hospitalized for additional diagnostic procedures, which included a contrast-enhanced CT scan to confirm the diagnosis. CT scans can also reveal a decrease in the volume of the affected lung segment. Additionally, ground-glass opacities may be observed, which could be associated with intra-alveolar hemorrhage and pulmonary congestion [13]. DSA represents the gold standard for the diagnosis of ASALLL. However, DSA is an invasive procedure and is expensive. CTA provides direct visualization of the abnormal blood supply artery's morphology and origin, as well as the development of the pulmonary artery in the affected lung segment. Also, the intuitive comprehension of the vascular anomalies can be improved by employing workstation-based multiplanar reformation (MPR) and volume rendering technique (VRT) to produce three-dimensional reconstructions of the anomalous artery and adjacent normal arteries. In this case, the primary goal following the patient's admission was to rule out a pulmonary space-occupying lesion. Therefore, only a single contrast-enhanced chest CT scan was conducted. The mediastinal window showed that the anomalous artery was located in the left lower lobe of the lung, where it originated from the left anterior wall of the descending aorta at the eighth thoracic vertebra. The artery exhibited a tortuous course and an S-shaped configuration, consistent with pulmonary artery CTA findings observed in most ASALLL patients [1, 23, 24]. The pulmonary arteries were found to be normally developed on both sides after post-processing with MPR and VRT. The lung window indicated that the left lower pulmonary bronchus was normally developed, while the involved lung exhibited congestive alterations. Other auxiliary examinations offer limited diagnostic information. Echocardiography is of great importance in evaluating pulmonary artery pressure, particularly regarding the presence of tricuspid and pulmonary regurgitation, the right-to-left ventricle diameter ratio, and the short-axis eccentricity index [25]. Pulmonary hypertension may develop in patients with ASALLL as a result of abnormal systemic artery shunting. An asymptomatic ASALLL patient was reported to have an elevated tricuspid regurgitation pressure gradient and mean pulmonary artery pressure by Sugihara et al.[6]. However, both abnormalities improved following treatment. The ultrasound examination verified that the patient did not exhibit any signs of pulmonary hypertension in this instance. In addition, the patient in our case exhibited slightly elevated levels of Red Blood Cell Count, Haemoglobin, Plateletcrit, and NSE, which had not been observed in previous cases. The correlation between these abnormal indicators and pulmonary vascular malformation remains unclear.

The treatment of ASALLL remains controversial. And there is no consensus exists on the necessity of treatment or the selection of surgical procedures. This pulmonary artery malformation has the potential to result in a range of complications, including pulmonary hypertension, hemoptysis, and heart failure, as evidenced by long-term follow-up data from a variety of case reports [6, 12, 26, 27]. A 28-year-old female patient with ASALLL died away as a result of massive hemoptysis during the observation period, as reported by Rubin et al. [28]. Ebihara et al. [29] performed a left lower lobe resection on a 65-year-old patient who had been diagnosed with ASALLL approximately 25 years earlier. During the subsequent long-term follow-up period of nearly 20 years, the patient developed thickened and calcified plaques in the abnormal systemic artery and also exhibited hypertension and hemoptysis. Machida et al. [12] followed up with an 80-year-old asymptomatic patient and observed that the diameter of the abnormal artery increased over 18 months. The findings suggest that ASALLL patients may be inclined to progress. However, patients may decline subsequent invasive treatment if they are asymptomatic or only experience minimal symptoms that can be alleviated with medication [22, 30]. Currently, there are limited long-term follow-up studies available for patients who do not receive treatment. Considering the long-term adverse effects of abnormal blood supply, timely intervention is imperative, regardless of the presence of symptoms. Treatment decisions should be based on the patient's age and the characteristics of their symptoms. Common treatment options include surgical intervention and interventional embolization. Surgical treatment has undergone a transformation from the traditional open thoracotomy to the modern minimally invasive thoracoscopy, including procedures such as lobectomy, segmentectomy, arterial ligation, and arterial anastomosis [19, 31–37]. In recent years, there has been a progressive increase in the use of embolization as a treatment option. Brühlmann et al. [36] were the first to effectively treat ASALLL by embolising the abnormal systemic artery with a coil in 1998. Embolisation accounted for 74% of the treatment cases from 2005 to 2022. In the selection of embolic materials, the coil remains the primary option, with the Amplazter Vascular Plug employed in certain instances [11, 34, 38]. A comprehensive review of the literature revealed no significant complications among patients who underwent embolization procedures. The potential complications primarily included post-embolization syndrome [34, 39] and chest discomfort (21%). A small number of patients experienced localized pulmonary infarction following surgery [31, 37–40], which typically resolves spontaneously in the short term. This may be attributable to the absence or degeneration of the

pulmonary artery in the affected segment, which could result in ischemia in that region of the lung following embolization. In this case, the patient was asymptomatic at the time of diagnosis. The echocardiogram, along with other tests, revealed no abnormalities. Given the highly invasive nature of the surgical procedure, spring coil embolization was performed. Embolization of the main trunk is analogous to endovascular ligation. It alleviates the pressure exerted by the abnormal side branch arteries on the pulmonary vascular bed due to elevated pressure, corrects the left-to-left shunt, reduces cardiac workload, and preserves as much normal pulmonary tissue as possible. The patient exhibited normal development of the pulmonary artery in the basal segment of the left lower lobe, and no complications such as pulmonary infarction occurred after embolization.

Conclusion

We treated an asymptomatic adult male patient with ASALLL using coil embolization. The patient had an anomalous branch arising from the thoracic aorta, supplying the basal segment of the left lower lobe, with normal development of the pulmonary arteries, lung parenchyma, and bronchi in the involved segment. The anomalous artery and distal branches were adequately embolized, and the patient experienced no postoperative complications. There are limited reports of embolization in asymptomatic patients. In order to determine whether intervention is required in these patients and to ascertain the subsequent impact of treatment, it is necessary to conduct a long-term follow-up and to examine additional cases.

Abbreviations

ASALLL	Anomalous systemic artery to the left lower lobe ()
NSE	Neuron-specific enolase
CT	Computed tomography
DSA	Digital subtraction angiography
CTA	Computed tomography angiography
MPR	Multiplanar reformation
VRT	Volume rendering technique

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Not applicable.

Author contributions

Meng Sun and Le Fang collected data of the patient and drafted the manuscript, Fangruyue Wang gathered the patient's medical records, Tianwei Wang designed the study and revised the manuscript. All authors have read and approved the final manuscript.

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Data availability

The data underlying this article will be shared on reasonable request to the corresponding author.

Declarations

Ethics approval and consent to participate

Since this is a case report, ethical review and approval were not required. The patient provided the written informed consent to participate in this study.

Consent for publication

Informed consent was obtained from the patient.

Competing interests

The authors declare no competing interests.

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