Letter to the Editor

Embolization via the brachial artery of an anomalous systemic artery supplying the left lower lung lobe: a case description

Mingyang Zhao[#], Xiaolong Zhang[#], Xi Zheng, Jieqiong Li, Ao Su, Yaxin Tuo, Enfu Du

Medical Imaging Center, Taihe Hospital, Hubei University of Medicine, Shiyan, China

[#]These authors contributed equally to this work and should be considered as co-first authors.

Correspondence to: Enfu Du, Master's degree. Medical Imaging Center, Taihe Hospital, Hubei University of Medicine, 30 South Renmin Road, Shiyan 442000, China. Email: def09120912@sina.com.

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Introduction

Anomalous systemic artery to the left lower lung lobe (ASALLL), also known as intrapulmonary sequestration (Pryce type I) (1), is a rare congenital pulmonary vascular abnormality. This disease occurs as a result of an aberrant artery originating from the descending aorta supplying the lower lung lobes. ASALLL occurs mostly in the lower left lobe (2) and rarely in the lower right lobe of the lung (3). The pulmonary arteries of the corresponding lobe may be completely absent (complete type) or partially present (incomplete type) (4). The pathophysiological basis of ASALLL is the congestion of the lung tissue supplied by the abnormal arteries. However, the lung parenchyma and bronchi of the affected lobe are not affected by this abnormality. ASALLL is generally not accompanied by clinical symptoms. In more severe cases, patients may present with sporadic or repeated coughing and hemoptysis. In addition, ASALLL may cause left atrial dilation, congestive heart failure caused by systemic hypertension, dyspnoea, chest pain, and lung infections (5).

The treatment of ASALLL is still controversial. The current treatment of ASALLL includes invasive surgical procedures such as lobectomy, vascular ligation, and vascular anastomosis. However, the high blood pressure within the thoracic aorta makes the surgical procedure challenging to perform and increases the risk of developing serious adverse effects such as hemoptysis, heart failure and rupture of the aberrant artery. Transarterial embolization (TAE) provide an alternative, noninvasive treatment for ASALLL. This technique involves the selective occlusion of a blood vessel via the image-guided introduction of emboli into a selected blood vessel.

This report presents the case of a woman diagnosed with incomplete type ASALLL, who was successfully treated with TAE via the brachial artery route. The clinical presentation, diagnostic workout, treatment procedure, and outcomes of this case are discussed in detail.

Case presentation

In 1999, a 42-year-old woman was diagnosed with ASALLL during a chest computed tomography (CT). She occasionally experienced mild hemoptysis and cough. However, the symptoms resolved on their own without special treatment. Progressive enlargement of the anomalous artery in the left lower lung lobe was noted during a physical examination performed in 2019. A CT of the thorax showed increased size and tortuosity of the blood vessels supplying the left lower lung lobe (*Figure 1A*). Subsequently, a computed tomography angiogram (CTA) of the aorta and pulmonary artery was performed. The volume reconstruction and multiplayer reconstruction images revealed an enlarged tortuous artery originating from the descending thoracic aorta perfusing into the left lower lobe (*Figure 1B,1C*). The CTA revealed a normal left pulmonary artery, except for a sparse lower branch (*Figure 1D*). The cardiac color doppler ultrasound showed a reduced diastolic function of the left atrium caused by an increased blood

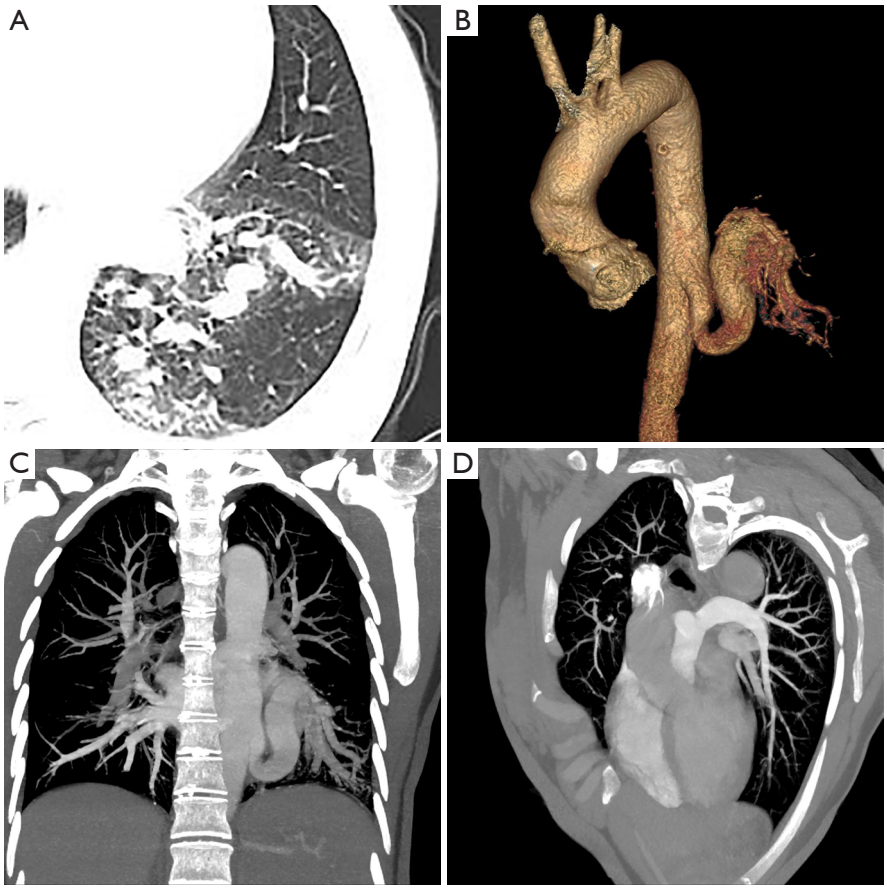

Figure 1 Preoperative images. (A) Chest CT demonstrates a significant increase in the blood vessel volume and tortuosity of the left lower lung lobe. (B,C) Aortic artery CTA demonstrating an enlarged tortuous artery originating from the descending thoracic aorta perfusing into the left lower lung lobe. (D) Pulmonary artery CTA showing a normal left pulmonary artery with a relatively sparse lower branch. CT, computed tomography; CTA, computed tomography angiogram.

flow from the left lower lobe to the left atrium. Based on the above examinations, the patient was diagnosed with a large anomalous artery. Since the angle of the anomalous artery in relation to the thoracic artery was too acute, a transarterial embolization (TAE) of the anomalous artery was performed via the left brachial artery to relieve the hypertension within the aberrant artery and allow the healthy pulmonary artery to perfuse into the left lower lobe normally. The final aim of this procedure was to resolve the patient's symptoms and limit the damage to the healthy lung tissue.

The Siemens Artis Zee Ceiling digital subtraction angiography (DSA) was used to guide the TAE procedure. Pulmonary arteriography was performed using a 5F pigtail catheter (Terumo, JP) via the right femoral vein. Subsequently, the left brachial artery was punctured, and the 5F catheter (MPA2, Cordis, USA) was guided toward the aberrant artery under angiographic image guidance.

The intraoperative pulmonary angiography confirmed that the left pulmonary artery was normal (*Figure 2A*). However, the thoracic aorta angiography revealed an aberrant artery supplying the left lower lung lobe. The anomalous systemic artery had an enlarged inner diameter measuring 12 mm (*Figure 2B*). Subsequently, an 8F-90 cm long sheath was positioned into the anomalous artery. The operators first tried to embolize the artery by releasing an 18 mm Amplatzer vascular plug (AVP). However, the plug was undersized with inadequate purchase and experienced distal embolization (*Figure 2C,2D*). Therefore, a larger AVP (20 mm AVP2, Shape Memory Medical, China) was deployed in appropriate proximal position resulting in occlusion of the anomalous systemic artery (*Figure 2E*).

After the TAE procedure, the patient reported chest

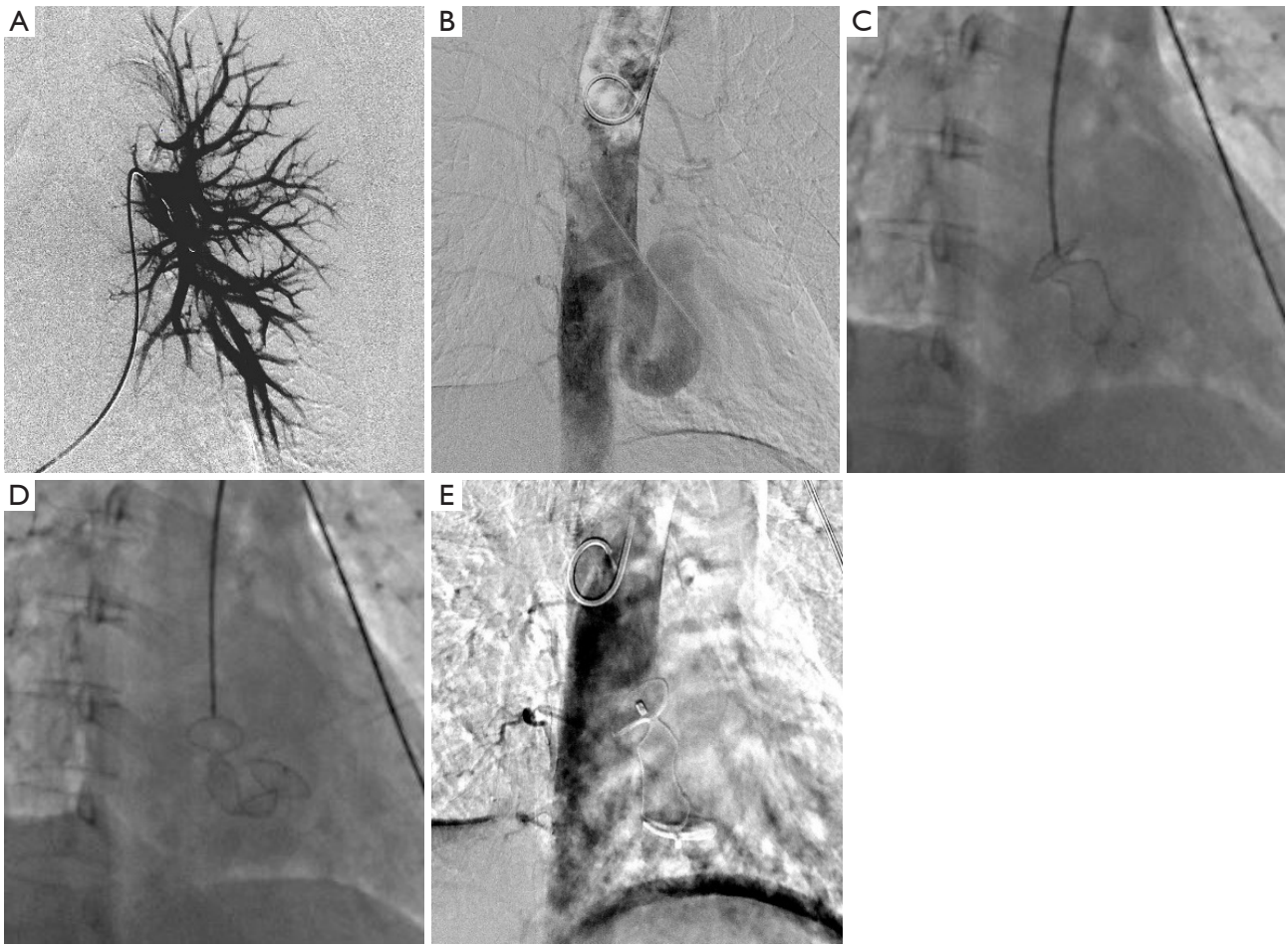

Figure 2 Intraoperative images. (A) Pulmonary angiography further confirmed that the left lower pulmonary artery was normal except for diminished branches in the left lower lobe. (B) The thoracic aorta angiography revealed an aberrant artery supplying the left lower lung lobe. (C,D) After the initial release of the 18mm AVP, displacement was observed on the DSA. (E) DSA acquired after the release of the 20 mm vascular plug. The new vascular plug was not displaced and successfully stopped the blood flow. AVP, Amplatzer vascular plug; DSA, digital subtraction angiography.

pain and mild fever, which was treated with an intravenous injection of 40 mg of dexamethasone. After about a week, the symptoms were gradually relieved. Three days after the TAE procedure, a chest CT was acquired. No obvious exudates were seen in the left lower lobe (*Figure 3*). The patient was followed up by telephone consultation. The patient reported that her cough, hemoptysis, and other symptoms improved significantly within six months post-operation. A contrast-enhanced CT of the chest and a CTA of the thoracic aorta were acquired after one month, three months, and six months post-operation. The postoperative CTA demonstrated expected thrombus in the anomalous systemic artery of the left lower lung lobe. Although the proximal end of the vascular plug had collapsed slightly, the position of the plug was still adequate. The CT demonstrated that the number of abnormal blood vessel branches in the lower lung lobe was substantially reduced, and the structure of the lung lobe was essentially restored. A cardiac color doppler ultrasound was also acquired three months post-operation. The ultrasound showed that the function of the left atrium was considerably restored.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case

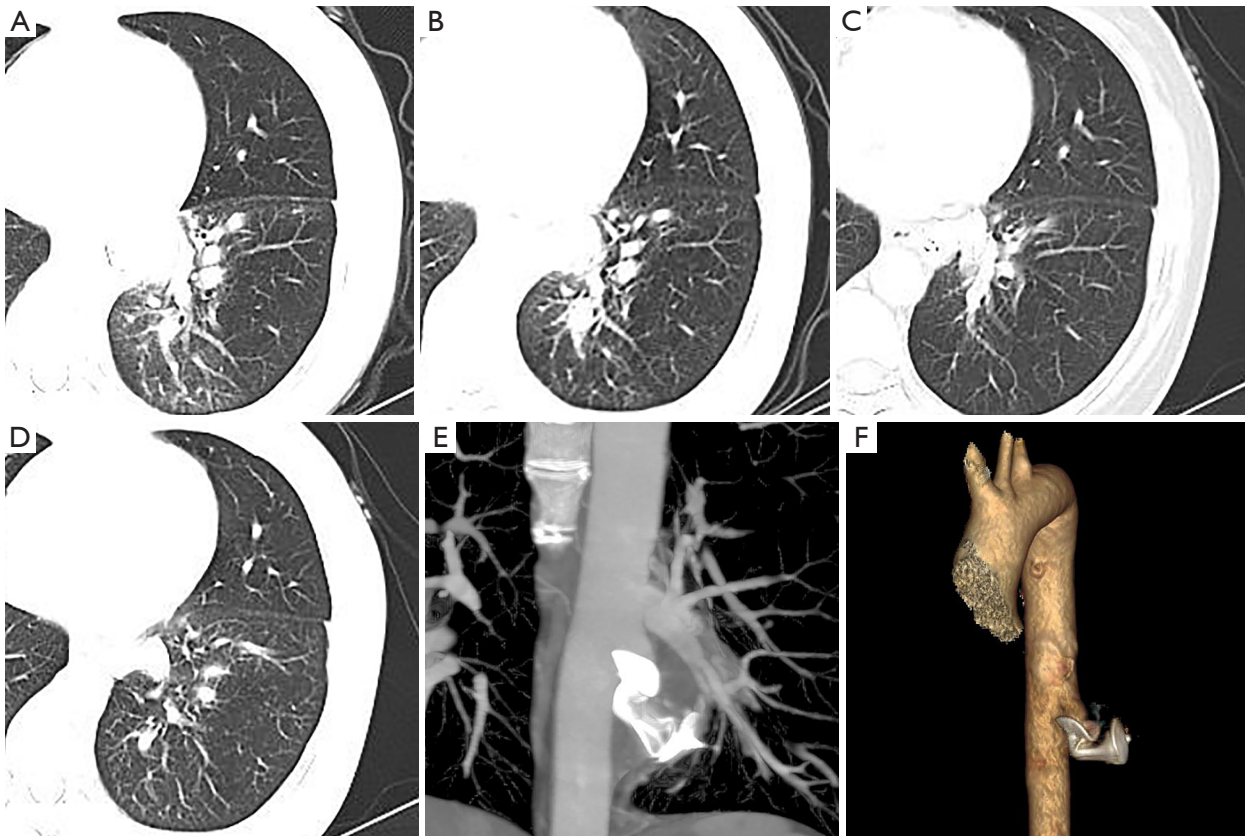

Figure 3 Postoperative images. (A,B) CT acquired after 3 days and 1 month postoperatively showed no obvious exudates in the left lower lung lobe and slightly sparse pulmonary arteries branching into the left lower lobe. (C,D) CT showing that the number of abnormal blood vessel branches was substantially reduced at three months and six months postoperatively. The structure of the lung lobes was completely restored six months after operation. (E) At one month post-operation, the CTA revealed the presence of an abnormal intra-body arterial thrombus, without any displacement of the vascular plug. (F) Postoperative CTA showing a slightly collapsed vascular plug at the proximal end. However, the position of the vascular plug six months after operation was still adequate. CT, computed tomography; CTA, computed tomography angiogram.

report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

In this manuscript, we present the case report of a patient diagnosed with ASALLL and treated with TAE via the brachial artery route.

ASALLL is a rare congenital vascular malformation of the pulmonary artery. At present, the cause of ASALLL has still not been determined. A study speculated that ASALLL results from a malformation within the original aortic branch that supplies the lung embryo before it develops into the embryonic pulmonary artery (6). ASALLL is often asymptomatic. This patient had no clinical symptoms for 20 years before the diagnosis of ASALLL. However, later on, the patient presented with shortness of breath and hemoptysis. The clinical manifestations and imaging characteristics of ASALLL are similar to those of intralobular pulmonary sequestration. The pathophysiological basis of ASALLL is the congestion of the lung tissue supplied by the abnormal arteries. The systemic blood pressure in ASALLL is higher than that of the pulmonary artery and may lead to intra-alveolar hemorrhage (7). If left untreated, the blood flow load on the left side of the heart may increase, thereby leading to an enlargement of the pulmonary vein and left atrium.

Eventually, the enlarged arteries and pulmonary veins compress the adjacent bronchi and cause tracheal stenosis.

Various imaging techniques can be used to diagnose ASALLL. DSA is currently considered the gold standard for the diagnosis of ASALLL. However, the procedure is invasive as it involves the insertion of a catheter into the artery. An alternative approach is the use of CTA. Studies have also shown that the diagnostic accuracy of CTA for detecting malformation within the pulmonary artery and thoracic aorta is similar to that of DSA (8,9). However, CTA is less invasive when compared with DSA as it does not require the insertion of a catheter into the femoral vein.

The current standard treatment of ASALLL includes lobectomy, vascular ligation, and vascular anastomosis of the anomalous artery to the remaining pulmonary artery. However, these surgical procedures carry considerable risk. As shown in this case, TAE can provide a safer and less invasive technique to occlude anomalous systemic arteries. Although complications such as chest pain, infection, and even pulmonary embolism may occur after TAE, the presence of a pulmonary artery supply makes TAE relatively safe in patients with the incomplete type of anomaly. Moreover, when compared with other surgical procedures, TAE provides faster recovery and has a lower rate of lung infection (3,10).

Anomalous systemic artery (ASA) usually originates from the left anterior wall of the lower thoracic descending aorta, and the trunk passes laterally downward, following a sigmoid shaped path (3). If the AVP delivery system enters the ASA through the femoral artery route, it can be very difficult control the release position of the AVP. In this case we report here, the angle between the anomalous artery and the thoracic aorta was too acute, causing the catheter tip to bend excessively when entering through the femoral artery route, thus increasing the difficulty of the operation. However, a change in the entry route can reduce the risk of bending the tip of the catheter thus increasing the success rate of TAE in ASALLL. Therefore, Canyigit *et al.* (11) proposed an alternative approach that involves guiding the AVP insertion via the right brachial artery. Another study successfully introduced a 6F-90-cm-long sheath into the ASA via the left radial artery (12). Moreover, in this case, the ASA was relatively thick, and therefore it was not possible to make use of a 6F-long sheath. Therefore, Under the guidance of the catheter and an exchange guide wire, an 8F-90-cm-long sheath was successfully introduced into the ASA and the AVP was successfully released. The selection of the appropriate AVP size is also very important to ensure that the blood flow does not displace the AVP. According to some scholars, it is recommended that the optimal size of the AVP should be at least 20% (preferably 30%) greater than the inner diameter of the target blood vessel (12). In this case, the inner diameter of the target artery was approximately 12 mm. An 18 mm AVP was initially inserted. The AVP diameter was selected so that its diameter was 1.5 times the size of the inner diameter of the target artery. However, the AVP quickly shifted to the distal end of the affected artery under the impact of blood flow. Therefore, a 20 mm AVP was used instead, which successfully blocked the ASA. The postoperative CTA showed a slight collapse in the proximal end of the AVP under the impact of blood flow. However, this did not affect the position of the AVP. A thrombus formation was also noted in the distal blood vessel, which further prevented the AVP from moving.

Several embolic materials, including coils, vascular occluders, detachable balloons, and AVPs, can be used to embolize a blood vessel. However, when compared with other emobolization techniques, coil embolization is simpler to perform. The problem is that the arterial anomaly in ASALLL is often large, and therefore a large number of coils would be required to stop the bleeding. On the other hand, the detachable balloons have poor vascular adhesion and eventually increase the risk of recanalization and operative time due to retraction. The AVP technique provides better vascular wall adhesion. Additionally, using an Amplatzer plug instead of multiple detachable coils is more cost-effective. Furthermore, the embolization effect is achieved via a single vascular occlude release. Nevertheless, as observed in this case, the diameter of the affected artery was too large and was not successfully occluded by an 18 mm AVP. For such cases, A study recommended using pushable coils combined with liquid embolic agents for embolization (13). Although the study showed that the spring coils could successfully embolize large anomalous arteries, the acute peripheral blood vessel occlusion caused by the liquid embolic agents increased the risk of developing pulmonary infarction and exudative changes. Since the anomalous artery was quite large in this case, the AVP method provided the best embolization technique. The postoperative follow-up examinations showed no evidence of pulmonary infarction or pulmonary infection. The patient's symptoms also resolved. The thrombus formation after the AVP embolization prevented any displacement of the AVP. Furthermore, the normal pulmonary arteries maintained the blood supply to the lungs, thereby avoiding pulmonary infarction or infection caused by acute terminal

blood vessel occlusion. The postoperative follow-up revealed that the structure of the lung lobes was also satisfactorily restored.

Conclusions

ASALLL is an extremely rare congenital pulmonary vascular malformation; thus, it is rarely encountered in clinical practice. In this case report, we present our perspective on the use of TAE via the brachial artery route to treat ASALLL. Compared with the femoral artery route, the brachial artery route facilitated the insertion of large AVPs into the affected blood vessel. Moreover, when compared with other embolic materials, the AVP technique can provide a simple embolization approach with fewer postoperative complications. Based on clinical experience of operators, when dealing with large anomalous systemic artery, the use of an AVP with a diameter 1.5 times that of the target blood vessel is recommended. This is because the fast blood flow rate in the thoracic aorta can increase the risk of displacement of the AVP, and using a larger diameter can help reduce this risk. Our technique successfully embolized the ASALLL, and the patient was asymptomatic at clinical follow-up at three year. However, long-term follow-up is required to further evaluate the efficacy of this technique.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-111/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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