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CASE REPORT

# Combined surgical therapy for pulmonary sequestration and aberrant artery from the abdominal aorta

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**Abstract**
Pulmonary sequestration with feeding vessels from the abdominal aorta is relatively rare. A 56-year-old woman with chronic left thoracic pain was referred to our hospital. Computed tomography showed multiple pulmonary cysts in the left lung and an aberrant artery from the abdominal aorta. She was diagnosed with pulmonary sequestration. She underwent embolization of the aberrant artery and wedge resection of the sequestrated lung under indocyanine green guidance. The surgical treatment combining preoperative embolization of the artery and intraoperative indocyanine green-guided lung resection might be safe and minimally invasive for patients with lung sequestrations accompanied by feeding vessels from the abdominal aorta.

**Keywords** Pulmonary sequestration · Embolization · Indocyanine green-guiding

## Introduction

Pulmonary sequestration (PS) is a congenital malformation that was first reported by Pryce in 1946 [1], and is defined as nonfunctional lung tissue that is separated from the normal lung. The descending thoracic aorta is the most common origin of the feeding artery as opposed to the abdominal aorta, which is the feeding artery in 6.9% of cases [2]. The standard treatment for PS is surgical resection via thoracotomy or video-assisted thoracic surgery (VATS). The standard procedure during surgery for PS is ligation or cutting of the aberrant artery combined with resection of the nonfunctional area of the lung parenchyma. However, few reports regarding treatment strategies for PS accompanied by aberrant arteries from the abdominal aorta have been published. We present the case of a patient with PS who had an aberrant artery from the abdominal aorta and was successfully treated through a combination of embolism and VATS wedge resection guided by indocyanine green (ICG).

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## Case

A 56-year-old woman was referred to our hospital because of chronic left chest pain for six months. Contrast-enhanced computed tomography (CT) showed multiple cystic lesions in the lower lobe of the left lung, which were not connected to the bronchi (Fig. 1a). Apparent boundaries were not observed between the normal lung and the lesion. The lesion was not supplied by the pulmonary artery but by an abnormal artery branching from the abdominal aorta, proximal to the celiac artery (Fig. 1b, c). Blood from the sequestrated lung drained into the left inferior pulmonary vein. Based on these radiological findings, the patient was diagnosed with left intralobar PS accompanied by a feeding artery from the abdominal aorta. Surgical treatment was chosen for this patient. Although ligation of the abnormal artery via laparotomy or laparoscopic surgery was considered, embolization of the artery was chosen to avoid abdominal surgery in addition to thoracic surgery. Briefly, a staged approach was chosen for the patient; embolization of the feeding artery by vascular surgeons under local anesthesia followed by resection of the sequestrated lung using VATS on the next day.

A 4F catheter was inserted into the feeding artery of the PS along the right femoral artery (Fig. 2a). Amplatzer vascular plug I (AVPI) was placed on the abdominal side of the diaphragm, and an 8 mm Ruby Coil was plugged in front of the AVPI with a peripheral occlusion device packing coil (Fig. 2b). Disruption of blood flow to the

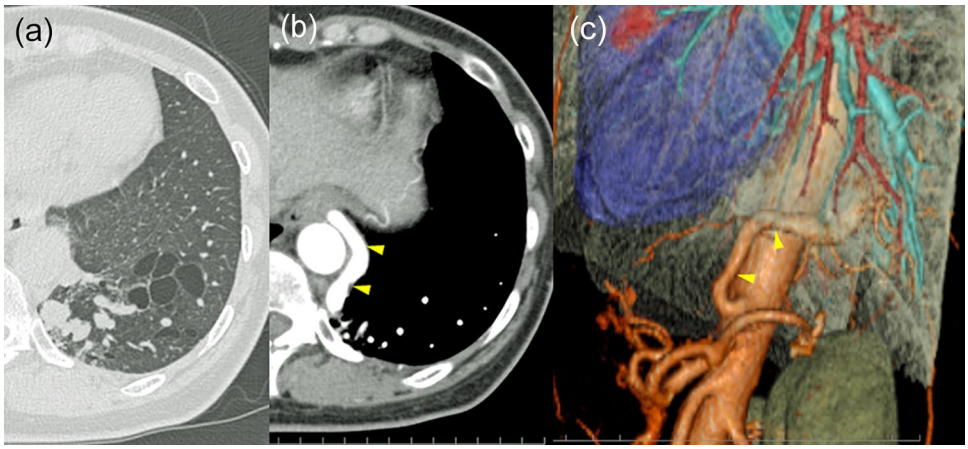


**Fig. 1** Contrast-enhanced computed tomography and 3-dimension angiography. **a** Computed tomography shows the cystic lesions in the left lower lobe. **b** Yellow arrowheads indicate an aberrant artery feeding the sequestrated lung. **c** Yellow arrowheads indicate an aberrant artery branching from the abdominal aorta under diaphragm

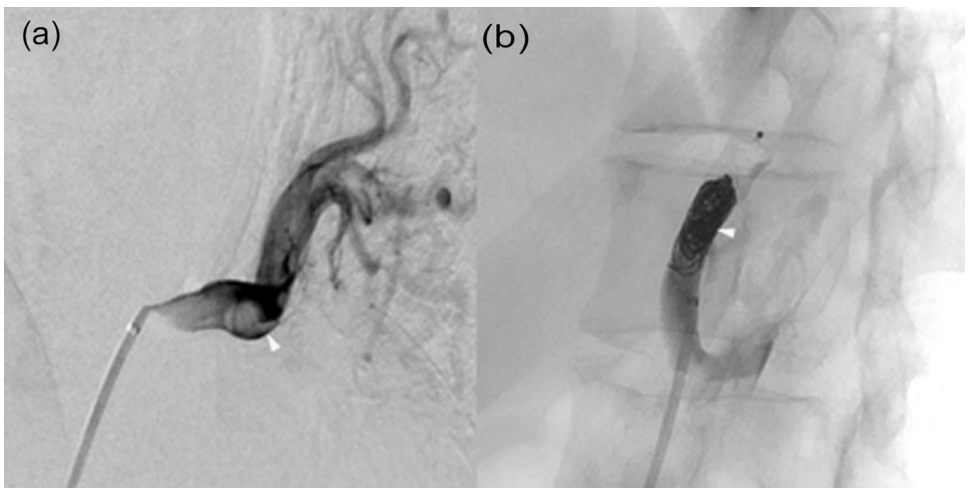


**Fig. 2** Angiography of the aberrant artery during embolization. **a** Angiography through the abdominal aorta shows the aberrant artery was branching from the abdominal aorta and feeding the left lower lung. **b** The aberrant artery was embolized with the vascular coil device (arrowhead)

sequestrated lung and preservation of the left subphrenic artery were confirmed. The patient's clinical course was uneventful after embolization. The day after embolization, pulmonary resection by VATS was performed. A 10 mm port was placed on the 8th intercostal space and a 30 mm-sized access window was opened in the 9th intercostal space. In the thoracic cavity, the fractional lung in the left lower lobe showed widespread adhesions to the diaphragm and descending aorta. The adhesions were detached, and it became clear that the abnormal artery from the abdominal cavity supplied the sequestrated lung through the aortic hiatus (Fig. 3a). The artery was dissected and excised with a stapling device 2 cm from the diaphragm. An aberrant vein from the sequestrated lung that drained into the inferior pulmonary vein was ligated and cut. Intravenous administration of 5-mg of indocyanine green (ICG) revealed a boundary between the normal and sequestrated lung (Fig. 3b). Wedge resection was performed using two staplers. The postoperative course was uneventful, and the patient was discharged on postoperative day 4. Six months after the surgery, she did not complain of pain, and CT showed no revascularization of the embolized artery.

## Discussion

We have presented a safe treatment technique using preoperative embolization of an aberrant artery from the abdominal aorta in a patient with intralobar PS. In addition, minimally invasive surgery was performed using ICG-guided excision.

Cases of intralobar PS with aberrant arteries from the abdominal aorta are rare. Fumimoto et al. reported a case of intralobar PS with an aberrant artery derived from the abdominal aorta treated by resection via VATS using stapler devices [3]. However, the risk of vascular injury and bleeding is relatively high because recurrent infections often cause abnormal vessels to become thicker and more fragile. When unexpected bleeding occurs, effective hemostasis may be difficult because of the high blood pressure of the aorta. If bleeding from injury to an aberrant artery occurs under the diaphragm, the bleeding points might be lost deep

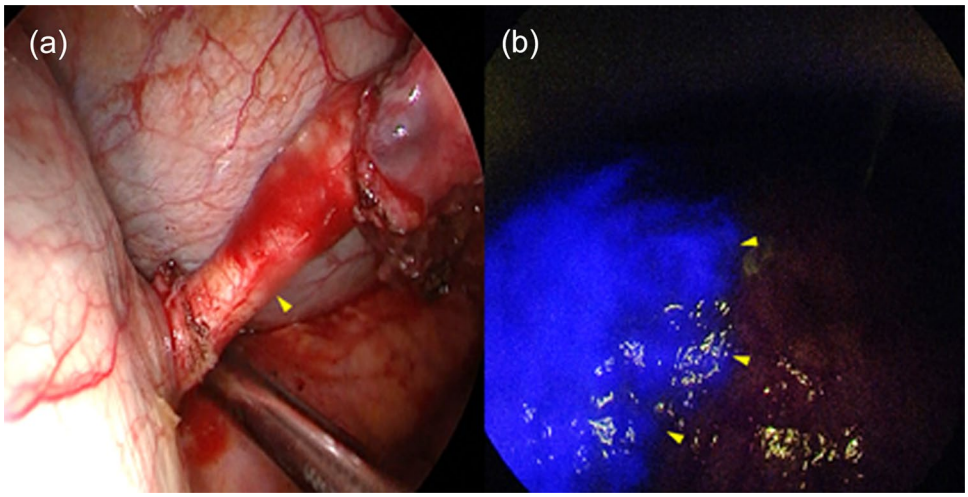


**Fig. 3** Intraoperative thoracoscopic findings. **a** Yellow arrowhead indicates an aberrant artery that passed through the aortic hiatus and supplied the left lower lung. **b** The indocyanine green-guided technique clearly showed the boundary between normal and sequestrated lung

into the diaphragmatic hiatus. In such a case, identifying the bleeding point and quickly performing hemostasis through the thoracic cavity would be very difficult. To avoid this, preoperative embolization of the aberrant artery from the abdominal aorta was useful for safe resection using a stapler during VATS. To our knowledge, only one case has been reported in which embolization of abnormal blood vessels from the abdominal aorta and safe removal of a sequestrated lung was performed [4]. In that case, embolization was performed just before lung resection in a hybrid operation room, while we performed embolization in the angiography room the day before lung resection. Our case demonstrates that even in facilities without special equipment such as a hybrid operating room, embolization for PS can be performed safely and easily in a normal angiography room.

Furthermore, we preserved the normal lung tissue as much as possible using the ICG-guided technique. ICG-guided lung resection for PS has been reported by Motono et al. [5]. ICG-guided wedge resection or segmentectomy is particularly useful for limited PS because it can preserve normal lungs as much as possible. In our case, resection of the feeding arteries and ligation of the vein enabled the boundary line between the normal lung and the PS to be clearly identified, and only the sequestrated lung was resected. Conversely, incomplete resection is a problem. Motohashi et al. argued that the cystic lung lesions recurred from the staple line after resection, and they aimed for complete resection of the lesions responsible for hyperinflated areas [6]. Therefore, accurate confirmation of the boundary between the PS and normal lung is important to prevent recurrence. For identification of the boundary between the PS and normal lung, an insufflation line is often used. However, in this patient, ICG was utilized because the PS lung showed severe emphysema and was not considered to be effective by air insufflation. Furthermore, ICG is cheap, widely available, and requires a relatively easy technique. On the contrary, the disadvantage of using ICG is that it can cause an allergic reaction in rare cases. The frequency of severe adverse events such as anaphylactic shock is reported as 0.05% [7]. Patients with an allergic predisposition should be carefully observed after ICG administration for early-, and late-phase allergic reactions.

## Conclusion

We present a safe and minimally invasive surgical treatment in a rare case of intralobar PS with a feeding artery from the abdominal aorta, by combining preoperative embolization and intraoperative ICG-guided resection. The combined use of preoperative embolization to reduce the risk of bleeding and ICG-guiding to define the boundary between the sequestrated and normal lung was very useful for the treatment of PS.

## Declarations

**Conflict of interest** None declared.

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