

## CASE REPORT

# Intralobar pulmonary sequestration originating from the intercostal arteries treated with surgical resection

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**Introduction**

Pulmonary sequestration is characterized by a rare congenital abnormality of the lower respiratory system arising from one or several aberrant systemic arteries, and accounts for 0.15–6.4% of all pulmonary malformations.<sup>1,2</sup> Typically, it is classified into two basic types: extralobar (25% of all cases), which is separated from the lung tissue by a separate lining of pleura; and intralobar (75% of all cases), which is embedded in the normal lung and shares common pleural investment.<sup>3,4</sup> In this case, final pathology revealed intralobar pulmonary sequestration.

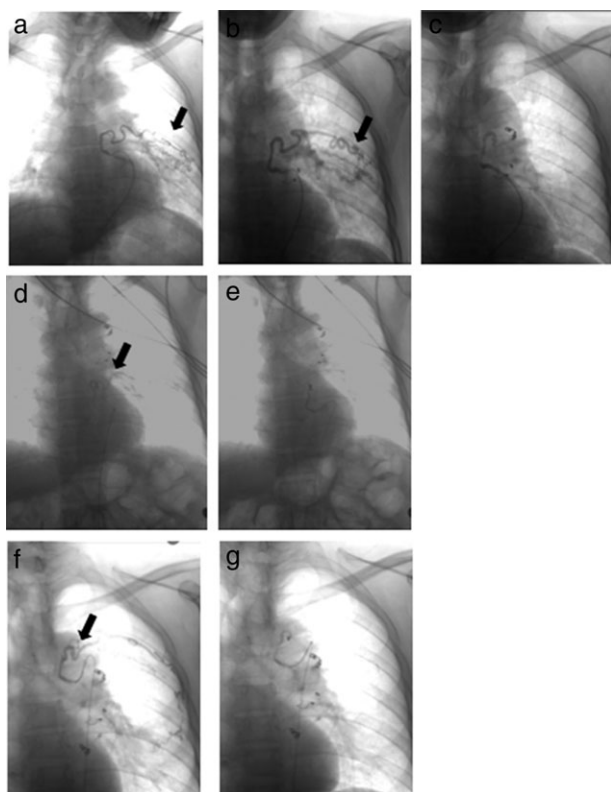
**Case Report**

A 56-year-old non-smoking male was admitted with repeated cough and massive hemoptysis that had lasted a

**Abstract**

Intralobar pulmonary sequestration originating from the intercostal arteries is rarely reported. Herein, we report an unusual case of a 56-year-old male patient with intralobar pulmonary sequestration supplied from the intercostal arteries on the left lower lobe who presented after a month of a repeated cough and massive hemoptysis. Although transcatheter arterial embolization was performed three times, the patient's symptoms were not relieved. A left lower lobectomy was performed with video-assisted thoracic surgery. At the six-month follow-up after surgery, the patient had recovered well without any hemoptysis. Therefore, surgical resection with lobectomy may be a better alternative to transcatheter arterial embolization for the treatment of intralobar pulmonary sequestrations arising from the intercostal arteries. To our knowledge, this is the second reported case of intralobar pulmonary sequestration arising from the intercostal arteries.

month. No other complaints or remarkable past medical history, including previous chest injury, were advised. The patient had been diagnosed a month earlier with pulmonary sequestration by chest computed tomography (CT) at another hospital, which indicated that the nutrient vessel was arising from the intercostal arteries. Transcatheter arterial embolization (TAE) had been performed three times at the previous hospital. After the right common femoral artery was accessed for digital angiography, some feeding arteries originating from the fifth and the sixth intercostal arteries were found, which supplied the abnormal lung tissue (Fig 1a–c). The aberrant arteries were then completely embolized with coils. After seven days, the second coil embolization was required because of continuous hemoptysis. Pre-embolization aortography demonstrated aberrant arterial blood supply from the seventh intercostal



**Figure 1** Transcatheter arterial embolization (TAE). (a–c) The first TAE: pre-embolization aortography shows the aberrant arterial blood supply from the (a) sixth and (b) fifth intercostal arteries; (c) the post-embolization aortography shows total occlusion of the aberrant arterial supply into the left lower lobe. (d–e) The second TAE was performed seven days after the first. The pre-embolization aortography shows the aberrant arterial blood supply from (d) the seventh intercostal artery and (e) the post-embolization aortography. (f–g) The third TAE was performed five days later. The pre-embolization aortography shows the aberrant arterial blood supply from (f) the fourth intercostal artery and (g) the post-embolization aortography.

artery, and the aberrant artery was completely occluded with coils once again (Fig 1d,e). The third embolization was performed five days later to block the aberrant arteries as a result of the recurrent hemoptysis. Pre-embolization aortography showed aberrant arterial blood supply from

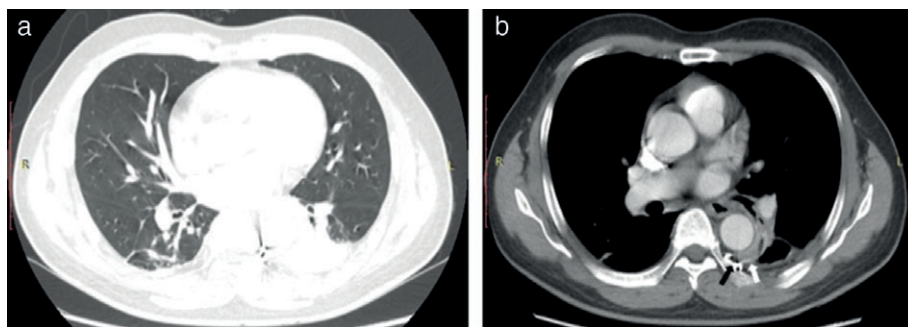
the fourth intercostal artery (Fig 1f,g). Although TAE was performed three times with platinum micro coils and gel foam before the patient presented to our department, the patient continued to experience hemoptysis measuring approximately 50 mL per day, indicating unsuccessful occlusion of the intercostal arteries.

We performed a post-embolization enhanced chest CT scan, which revealed an irregular margin of a pulmonary mass on the left lower lobe with a feeding vessel originating from the intercostal arteries, particularly on the basal segments (Fig 2). Some metal micro coils were also observed on the CT scan. The radiological findings indicated pulmonary sequestration. A general left lower lobectomy was performed by video-assisted thoracic surgery (VATS) because of the repeated cough and massive hemoptysis. A mass in the base of the left lung was resected during surgery, and aberrant arteries woven into vascular vines originating from multiple intercostal spaces leading to the abnormal lung tissue were observed (Fig 3a, b). The aberrant arteries were carefully separated from the surrounding adhesion and then divided using an endothoracic stapling device. Standard thoracoscopic lobectomy was then performed and the tissue was sent for histopathology. The final histopathology report suggested an intrapulmonary sequestration (Fig 3c,d). The patient was discharged on the fifth postoperative day. After six months of follow-up, the patient completely recovered without any complaint.

Written informed consent was obtained from the patient for the publication of this case report and the accompanying images.

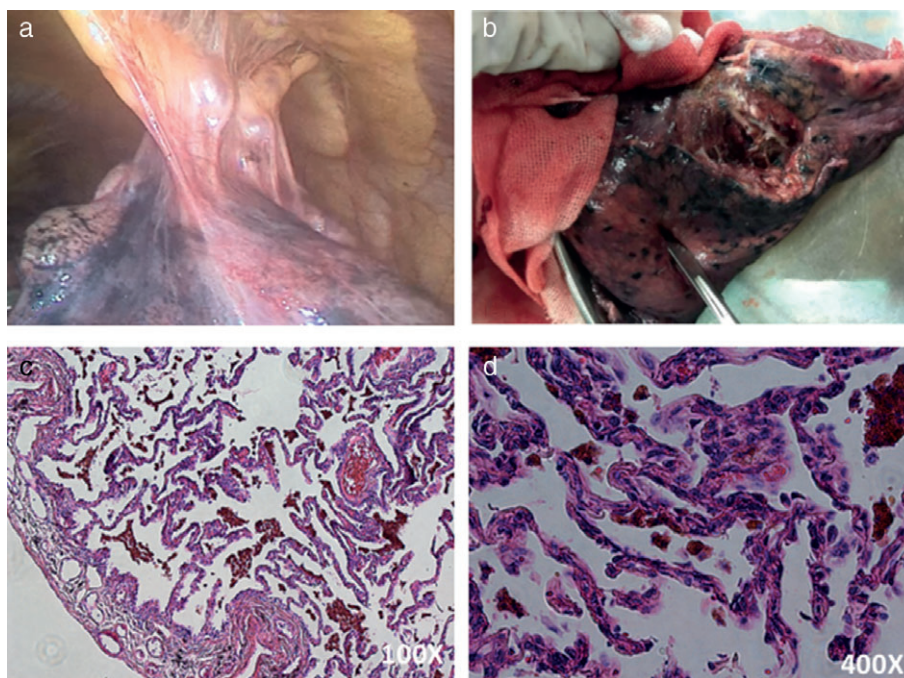
## Discussion

It is theorized that the pathogenesis of sequestration is that early embryonic splanchnic vessels develop into systemic vessels. The sequestration subsequently receives systemic arterial supply instead of the normal pulmonary arterial supply.<sup>4,5</sup> Intralobar pulmonary sequestrations receive anomalous systemic arterial supply predominantly deriving from the descending thoracic aorta (72%), abdominal aorta (21%), and, rarely, the intercostal arteries (3%).<sup>6</sup> The



**Figure 2** Enhanced chest computed tomography (CT) scan after three embolization procedures. (a) Lung window axial thorax CT image shows a pulmonary mass on the left lower lobe with irregular margins. (b) A contrast-enhanced view shows a feeding vessel (white arrow) originating from the intercostal artery draining into the lesion. Embolization material is observed in the CT (black arrow).

**Figure 3** Images of intraoperative left lung and pathology. (a) Intraoperative thoracoscopic images show the aberrant vessels originating from the intercostal arteries and leading to the mass. (b) The gross specimen reveals a pulmonary mass with some tiny central cystic-type spaces on the left lower lobe, consistent with pulmonary sequestration. (c–d) The pathology revealed intralobar pulmonary sequestration by hematoxylin and eosin staining.



majority of intralobar sequestrations are drained by normal pulmonary veins into the left atrium.<sup>7</sup> In this case, the feeding artery originates from the intercostal arteries. It is rare and unusual. A study of 105 cases revealed that 74 were derived from the descending thoracic aorta, 25 from the abdominal aorta, 5 from the intercostal arteries, and 1 from the aortic arch.<sup>5</sup>

Patients can present with an incidental pulmonary lesion on imaging while asymptomatic.<sup>6</sup> However, patients may present with cough, fever, recurrent pneumonia, chest pain, and in a small number of cases, hemoptysis. A study of 73 cases revealed presenting symptoms of pneumonia ( $n = 11$ ), pneumothorax ( $n = 2$ ), pneumomediastinum ( $n = 1$ ), and respiratory distress ( $n = 6$ ). Fifty-three patients were asymptomatic.<sup>8</sup>

Surgical resection is the classical therapeutic approach for patients with pulmonary sequestration in order to prevent recurrent pulmonary infections, massive hemoptysis, or other symptoms, but its value is debatable in asymptomatic patients.<sup>9</sup> Recently, improved access to interventional radiology treatment coupled with advances in the coil embolization technique has been reported as a minimally invasive surgical alternative to lobectomy<sup>2,4,10,11</sup> However, data of endovascular embolization in symptomatic adult patients is sparse and long-term efficacy and safety have not been reported. Furthermore, very few studies have compared the results of embolization and surgical resection and reported which treatment is better in adult patients.<sup>12</sup> Some authors believe that embolization can occlude the arterial supply, leading to sequestration

infarction, and thus is an alternative treatment method to conventional surgical resection.<sup>10</sup> As such, embolization is thought by some to be superior to surgery as it minimizes complications and shortens hospital stay.<sup>13</sup>

An adverse effect of embolization is the distant migration of embolization products resulting in the embolization of non-targeted arteries.<sup>13</sup> We present a case of a 56-year-old patient affected by intralobar pulmonary sequestration. He required a second coil embolization seven days after the first because of continuous hemoptysis and residual systemic flow, as seen in angiography. The third embolization was performed five days later to block the aberrant arteries, because of the recurrent hemoptysis. The necessity of repeat embolization indicates that if there are multiple abnormal arteries to supply the lesion, it is better use a surgical solution.

Surgical treatment should be considered in cases of complete or partial failure of endovascular treatment. Incomplete embolization may result from distal occlusion of the feeding artery with possible subsequent opening of collateral supply. After incomplete embolization, recurrence of symptoms and infection may occur.<sup>13</sup> We believe that surgical treatment is feasible, efficacious, and a safe alternative to TAE, which carries a risk of recurrent infection and hemoptysis, a low rate of regression, and complications after embolization, and to exclude other pathology. Our case shows that VATS offers an important alternative to the open approach for pulmonary sequestration with minimum surgical trauma morbidity, postoperative pain, and hospitalization. The feasibility of VATS resection has been

shown in many case reports.<sup>1,5,8</sup> Special attention needs to be paid to the identification of aberrant arteries because these need to be identified before they are ligated and divided. Thoracoscopic treatment of the aberrant artery of pulmonary sequestration is feasible in experienced hands. The aberrant systemic arteries can be freed and dissected safely despite any inflammatory changes as a result of recurrent infection or when arteries are hidden in adhesive tissue. The conversion rate to thoracotomy is low. In our case, the VATS procedure was performed under single lung ventilation in a full right lateral position. Intraoperative thoracoscopic exploration revealed a mass in the base of the left lung and aberrant arteries woven into vascular vines originating from multiple intercostal spaces and leading to the abnormal lung tissue. We carefully separated the aberrant artery adhesion from the surrounding tissues and used an endothoracic stapling device to divide them. A standard thoracoscopic lobectomy was then performed. The six-month follow-up showed that the efficacy of VATS was superior to embolization because the patient did not experience postoperative complications.

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## Disclosure

No authors report any conflict of interest.

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