Clinical Case Report

Medicine®

OPEN

Endovascular treatment of pulmonary sequestration with thoracic endograft

Two case reports

Yu Chen, MD, Bao Liu, MD, Jiang Shao, MD, Duan Liu, MD, Yuehong Zheng, MD*

Abstract

Introduction: Pulmonary sequestration (PS) is a rare congenital lung malformation. The classical treatment of the disease has been conventional surgery with resection of abnormal parenchyma. Recently, the endovascular embolization has been proposed for the treatment of this disease. Here we present 2 cases of PS successfully treated with thoracic endograft.

Patient concerns: Two patients with abnormal consolidation in the left lower lobe were admitted in our hospital.

Diagnosis: Chest computed tomography angiography (CTA) showed abnormal consolidation in the left lower lobe, which received systemic blood supply from the descending aorta in both patients. So the diagnosis of PS was confirmed.

Interventions: Endovascular treatment with thoracic endograft was successfully performed.

Outcomes: The patients recovered well and were completely free of symptoms. And the CTA follow-up showed the abnormal pulmonary parenchyma shrunk significantly.

Conclusions: Endovascular treatment with thoracic endograft is a promising treatment option for PS.

Abbreviations: CT = computed tomography, CTA = computed tomography angiography, ELS = extralobar sequestration, ILS = intralobar sequestration, PS = pulmonary sequestration.

Keywords: embolism, endovascular treatment, pulmonary sequestration

1. Introduction

Pulmonary sequestration (PS) is a rare congenital malformation of the lower respiratory tract, and the standard treatment of the disease has been conventional surgery with resection of abnormal parenchyma. However, during pulmonary resection, inadvertent injury of the aberrant artery can cause a life-threatening hemorrhage. So endovascular embolization has been proposed for the treatment of this disease. But if the embolization is incomplete, the infection would recurred. And sometimes embolization would cause severe parenchyma necrosis. This article presents 2 cases of PS which were successfully treated with thoracic endograft, and brief review of the clinical features, diagnostic strategies, and current management of the PS.

2. Clinical cases

2.1. Case 1

A 36-year-old man was admitted in the hospital for recurrent pneumonia. He had been treated with antibiotics for 1 week and the symptom was released. The thoracic computed tomography (CT) revealed an area of consolidation within the left lower lobe of the lung. After treatment of antibiotics, the pneumonia was cured, but the CT showed the mass was still there. Computed tomography angiography (CTA) identified a tortuous artery coming from the descending thoracic aorta and supplying the abnormal pulmonary parenchyma (Fig. 1), so the diagnosis of PS was confirmed. One week later there was no sign of infection in the patient, and we performed endovascular treatment with thoracic endograft. Under general anesthesia, a short 8F introducer sheath (Cordis, Miami, FL) was put in place via the right transfemoral route and 2 Perclose ProGlide sutures (Abbott, Chicago, IL) were deployed. Angiographic evaluation confirmed the presence of the aberrant nutrient arteries (Fig. 2). Then the introducer sheath was moved and a thoracic endograft (Endurant 28-28-70 mm, Medtronic, Minneapolis, MN) was put into the thoracic aorta and covered the orifice of the abnormal artery. The final angiogram confirmed that the aberrant arteries were successfully occluded (Fig. 3). Then the preformed knots were tightened and manual pressure applied. After the operation the patient had taken oral antibiotics for 1 week to avoid infection. No incident or complication occurred during the procedure.

Editor: N/A.

Ethical approval to report these 2 cases was obtained from Ethic committee, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences. And written informed consent was obtained from the patients for their anonymized information to be published in this article.

The authors have no conflicts of interest to disclose.

Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences, Beijing, China.

** Correspondence: Yuehong Zheng, Department of Vascular Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences, Beijing 100730, China (e-mail: yuehongzheng@yahoo.com).*

Medicine (2019) 98:31(e16666)

Received: 22 January 2019 / Received in final form: 29 May 2019 / Accepted: 8 July 2019

http://dx.doi.org/10.1097/MD.0000000000016666

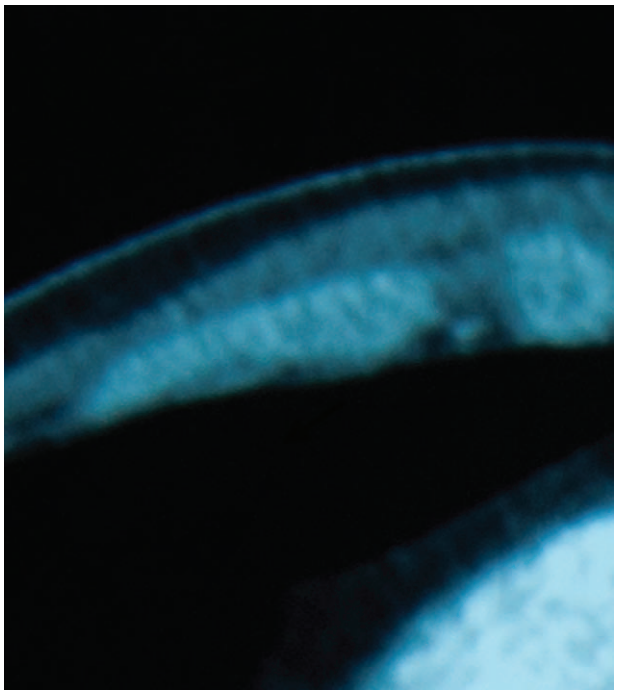

Figure 1. CTA revealed the anomalous inflow vessel arising from the thoracic aorta and supplying abnormal pulmonary parenchyma (Marked by black arrow). CTA = computed tomography angiography.

Postoperatively the patient recovered well, and at follow-up after 3 months, the patient did not suffer pneumonia again. The CTA showed no sign of the abnormal nutrient arteries (Fig. 4) and the abnormal pulmonary parenchyma shrunk significantly (Fig. 5). One year later, the patient did not suffered from recurrent pneumonia and the CTA showed the abnormal pulmonary parenchyma did not get enlarged (Fig. 6).

2.2. Case 2

A 37-year-old male patient was admitted in the hospital for chest distress and chest pain on inspiration. Prior workup for acute coronary syndrome was negative, and echocardiography revealed no evidence of heart disease or congenital malformations. CTA identified increased lung marking in the left lower lobe and a tortuous artery coming from the descending thoracic aorta, supplying the left lower lobe, so PS was diagnosed. Because the aberrant nutrient artery was extremely big and tortuous, it is difficult to control or embolize them. We decided to perform endovascular treatment with thoracic endograft. This time we used the covered stent (Ankura 28-28-80, Lifetech, Shenzhen, China) to cover the orifice of the aberrant nutrient artery and the final angiography confirmed the abnormal artery was totally occluded. No incident or complication occurred during the procedure. The patient uneventfully discharged home on post-operation day 3. And he also took antibiotics orally for 1 week after the operation. At follow-up after 3 months, the patient was completely free of symptoms.

3. Discussion

PS is defined as nonfunctioning mass of lung tissue that lacks normal communication with the tracheobronchial tree and

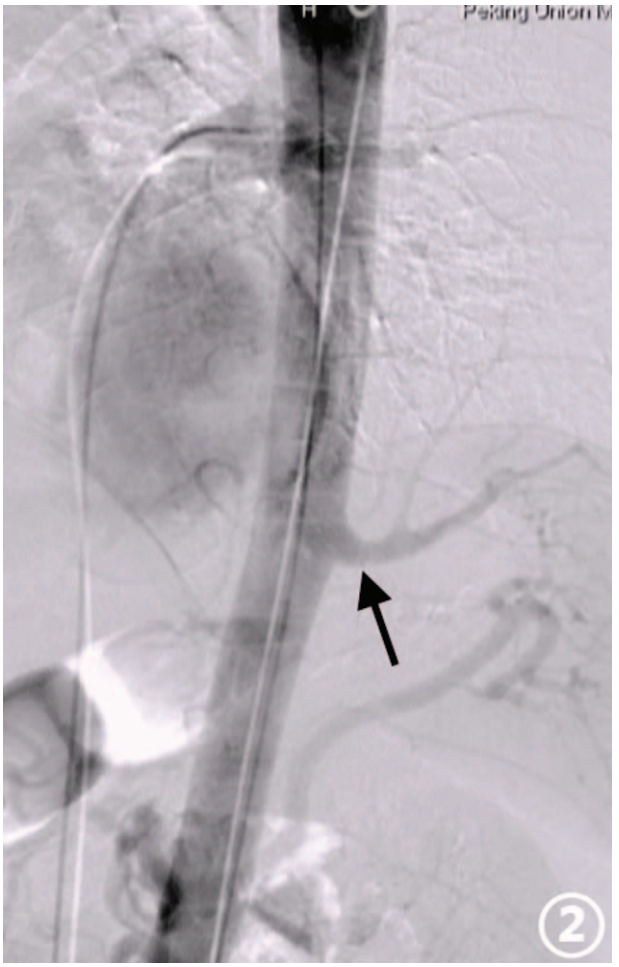

Figure 2. The initial diagnostic aortography confirmed an abnormal artery originated from the thoracic aorta. (Marked by black arrow).

receives its arterial blood supply from the systemic circulation.[1] PS is a rare malformation of the lower respiratory tract which comprised 0.15% to 6.4% of all congenital pulmonary anomaly.[2] PS can be classified as intralobar sequestration (ILS) or extralobar sequestration (ELS) according to its relationship with the visceral pleura. ILS, accounting for about 75% of PS, is located within the normal lung parenchyma and shares the visceral pleura of the parent lobe of the lung; while ELS is located outside the normal lung and has its own visceral pleura. PS receives systemic artery supply from the descending thoracic aorta, abdominal aorta, the celiac trunk, intercostal arteries, and so on.[3] ILS has normal pulmonary venous return, while ELS is associated with aberrant pulmonary venous drainage to the azygos system. The majority of patients are asymptomatic, however, some patients suffer from some clinical symptom, such as recurrent pulmonary infection, cardiac failure due to left-to-right shunt and hemoptysis.[4] Almost 97% of PS cases were located in the lower lobe, and PS in the left lower lobe is 2 to 3 times more common than that in the right lower lobe.[5]

The diagnosis of PS is based on imaging and identifying the systemic arterial supply. Pulmonary angiography is thought to be the gold standard for diagnosis; however, in recent years, several studies have shown that less invasive imaging techniques such as CTA and magnetic resonance angiography may be equally effective and safer

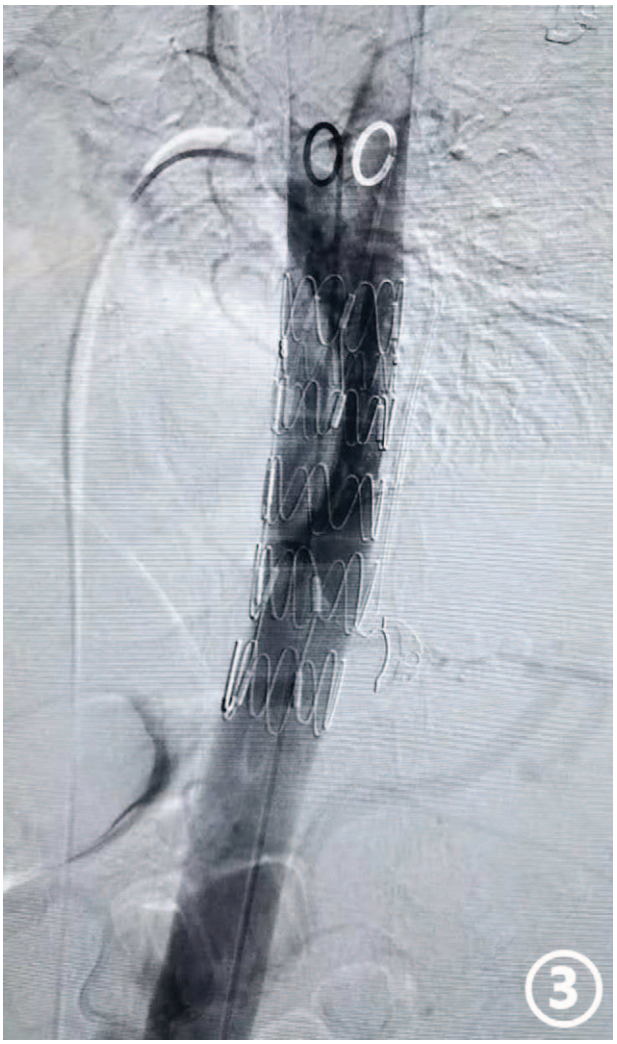

Figure 3. After the thoracic endograft was deployed, the abnormal artery was totally occluded.

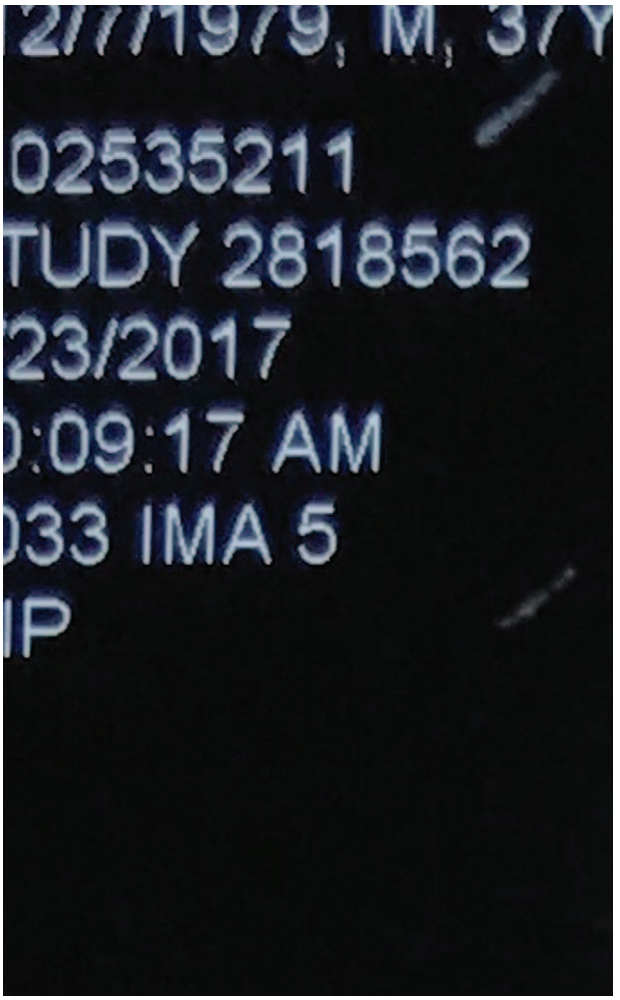

Figure 4. Three months later, the CTA showed no sign of the abnormal artery and the abnormal pulmonary parenchyma shrunk significantly. CTA= computed tomography angiography.

alternatives. Now, thoracic CTA has been the most useful test in the evaluation of patients suspected PS, and it can demonstrate both the bronchial and vascular anomaly of the lung.[6]

The standard treatment of PS is resection of the segment or lobe that contains the sequestered tissue by thoracotomy or thoracoscopic approach. However, inadvertent injury of the aberrant artery can cause severe and potentially life-threatening hemorrhage. The aberrant artery needs to be identified and controlled early during the surgical resection, but the artery could be friable due to chronic inflammation, which make it difficult to control.[7] Preoperative embolization has been proposed to prevent intraoperative bleeding. It can obliterate the major arterial supply to the sequestrated lung tissue, thus making the surgical procedure much easier and safer.

Endovascular embolization of PS has been reported as a safe alternative to surgery since 1998[8] and the data of the treatment shows satisfactory results. Some cases in children even showed good results with complete lesion regression.[9] Major concerns about endovascular embolization include possible incomplete occlusion of arterial supply, resulting from distal embolization of feeding vessel with subsequent opening of collateral supply. This may lead to evolution of the sequestered tissue and possible recurrence of symptoms. On the other hand, sometimes the embolization could block almost all the blood to the sequestrated parenchyma, which could cause massive necrosis and pulmonary abscess.[10] Distant migration of embolization material, resulting in embolization of nontargeted arteries has also been reported.[11]

Endovascular treatment with thoracic endograft is another option.[12] This kind of treatment is easy to achieve, especially in the cases with big and tortuous nutrient artery. But just like embolism, because no resection of the PS was performed, there is a possible risk of symptoms recurrence or lesions enlarged. Therefore, it is important to wait for late follow-up results.

Because of the significant improvement of symptoms, there was no need for surgery. But the patients were informed that surgical treatment could be considered in case of clinical symptoms recurrence or lesions enlarged. Long term follow-up was required to determine whether surgical resection of PS was needed.

Nowadays no treatment guidelines have been established. Some studies drew conclusions that endovascular technique was considered for patients when pulmonary lesions were small-sized (<3 cm).[13] Due to a retrospective study and a limited number of cases, we cannot conclude the specific size of the sequestration as a basis for treatment option in our study. Prospective studies and

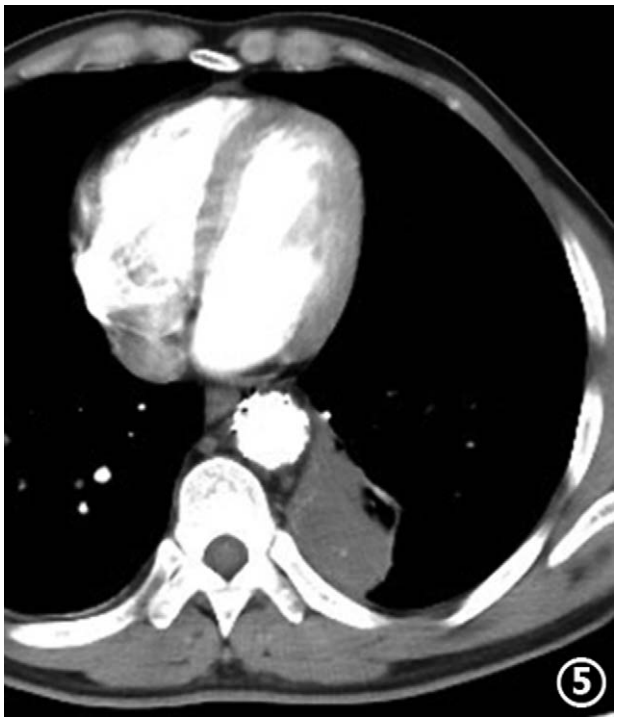

Figure 5. Three months later, the CTA showed no sign of the abnormal artery and the abnormal pulmonary parenchyma shrunk significantly. CTA= computed tomography angiography.

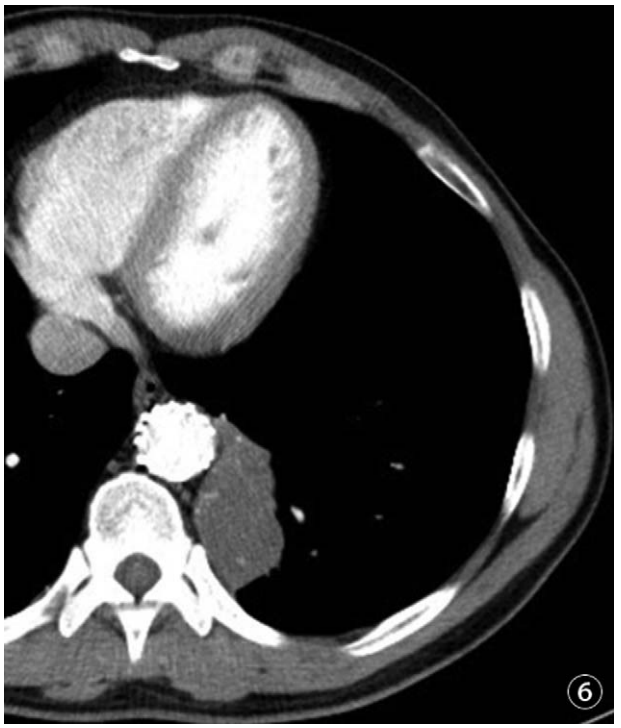

Figure 6. One year later, the abnormal pulmonary parenchyma was still there and did not change a lot.

large sample are needed to establish criteria for treatment options based on lesion size.

Major concern about the thoracic endograft is the risk of stent infection. Curses et al think that an already infected sequestration should not be managed with endovascular treatment.[14] The first case had pneumonia, and after the inflammation was controlled, he was treated with aortic covered stent. Up till now, the patient does not show any sign of infection of the covered stent. So we think it is safe to use thoracic endograft in PS if the pneumonia was cured. But concerning the disastrous sequence of the stent infection, we suggest the treatment should be performed at least 1 week after the pneumonia is cured, and the patient should be treated with antibiotics for at least 1 week after the endovascular treatment. And if the infection of the sequestrated tissue occur, the resection of the PS should be performed without delay.

We believe that endovascular treatment with thoracic endograft is a promising treatment option for PS, but more evidence-based conclusions are needed to certify its long-term safety and efficacy.

Author contributions

Data curation: Duan Liu.
Investigation: Bao Liu, Jiang Shao.
Writing – original draft: Yu Chen.
Writing – review and editing: Yuehong Zheng.

References

[1] Landing BH, Dixon LG. Congenital malformations and genetic disorders of the respiratory tract (larynx, trachea, bronchi, and lungs). Am Rev Respir Dis 1979;120:151–85.
[2] Ferris EJ, Smith PL, Mitza FH, et al. Intralobar pulmonary sequestration: value of aortography and pulmonary arteriography. Cardiovasc Intervent Radiol 1981;4:17–23.
[3] Avsenik J, Stupnik T, Popovic P. Endovascular embolization prior to surgical resection of symptomatic intralobar pulmonary sequestration in an adult. Eur J Radiol Open 2015;9:12–5.
[4] Mautone M, Naidoo P. A case of systemic arterial supply to the right lower lobe of the lung: imaging findings and review of the literature. J Radiol Case Rep 2014;8:9–15.
[5] Sun X, Xiao Y. Pulmonary sequestration in adult patients: a retrospective study. Eur J Cardiothorac Surg 2015;48:279–82.
[6] Yue SW, Guo H, Zhang YG, et al. The clinical value of computer tomographic angiography for the diagnosis and therapeutic planning of patients with pulmonary sequestration. Eur J Cardiothorac Surg 2013;43:946–51.
[7] Goto T, Toya K, Wakaki M, et al. Resection of intralobar pulmonary sequestration after coil embolization of aberrant arteries: report of a case. Surg Today 2013;43:923–5.
[8] Park ST, Yoon CH, Sung KB, et al. Pulmonary sequestration in a newborn infant: treatment with arterial embolization. J Vasc Interv Radiol 1998;9:648–50.
[9] Curros F, Chigot V, Emond S, et al. Role of embolisation in the treatment of bronchopulmonary sequestration. Pediatr Radiol 2000;30:769–73.
[10] Marine LM, Valdes FE, Mertens RM, et al. Endovascular treatment of symptomatic pulmonary sequestration. Ann Vasc Surg 2011;25:696.e11–5.
[11] Leoncini G, Rossi UG, Ferro C, et al. Endovascular treatment of pulmonary sequestration in adults using Amplatzer® vascular plugs. Interact Cardiovasc Thorac Surg 2011;12:98–100.
[12] Wilder FG, Minasyan SZ. Thoracic stent graft accompanied by coil embolization for pulmonary sequestration. Innovations (Phila) 2019;14:168–73.
[13] Zhang SX, Wang HD, Yang K, et al. Retrospective review of the diagnosis and treatment of pulmonary sequestration in 28 patients: surgery or endovascular techniques? J Thorac Dis 2017;9:5153–60.
[14] Curros F, Chigot V, Emond S, et al. Role of embolisation in the treatment of pulmonary sequestration. Pediatr Radiol 2000;30:769–73.