Aberrant Feeding Artery Mutilation Treatment of Pulmonary Sequestration: A Case Report

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Case report

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Abstract

Background: Pulmonary sequestration (PS) is a rare congenital pulmonary pulmonary malformation. In this study, we reported a case of PS treated with aberrant feeding artery mutilation in our center.

Case Presentation: A 29-year-old female patient presented to our center in March 2020 due to repeated hemoptysis for more than 15 years, she received CTA examination which showed an aberrant feeding artery from the thoracic aorta into the left lower lobe of the lung and was therefore diagnosed with left lower lung pulmonary sequestration. Given that she complained with no infectious symptoms, she was treated with thoracoscopic feeding artery mutilation in June 2020. She and experienced an uneventful postoperative course and was discharged 2 days postoperatively. On the postoperative follow-up of 4 months, this patient's physical condition was excellent and free of any symptoms such as hemoptysis or cough.

Conclusion: Surgery is still the first choose for the treatment of PS. When pulmonary lesion is small-sized, for PS patients with non-infectious symptoms, aberrant feeding artery mutilation can be attempted as an alternative to intravascular technology for the treatment of PS. Future large studies are required to further verify its long-term efficacy and scope of application.

Introduction

Pulmonary sequestration (PS) is a rare congenital respiratory malformation[1], The pathogenic mechanism of pulmonary sequestration is still unknown. Several etiological hypotheses of PS have been proposed[1, 2], the most widely accepted of which suggests that PS results from the formation of an accessory lung bud caudal to the normal lung buds [1–3], and studies show that Extralobar pulmonary sequestrations were likely to regress spontaneously[4, 5]. PS is anatomically characterized by the presence of feeding arteries and non-functional lung tissue, and PS is more likely to occur in lower lungs. Due to the atypical clinical symptoms of PS patients, it is difficult to differentiate with a variety of other pulmonary diseases[2]. The treatment strategies for PS are controversial.

Case Presentation

A 29-year-old female patient presented to our center in March 2020 due to repeated hemoptysis for more than 15 years. In the past 15 years, she had visited hospitals for many times due to repeated hemoptysis, and the symptoms of hemoptysis were relieved after symptomatic support treatment such as infusion and hemostasis. In March 2020, the symptoms of hemoptysis were worse than before, then she presented to our center and she received CTA examination which showed an aberrant feeding artery from the thoracic aorta into the left lower lobe of the lung and therefore diagnosed with left lower lung lobe intralabor PS(Fig. 1a). She has no special medical history and no obvious abnormality in physical examination. In June 2020, she received uniportal video-assisted thoracoscopic feeding artery mutilation, through the 5th intercostal utility incision. During the operation, the aberrant feeding artery about 10 mm
in diameter was found in the left lower lobe near the lower pulmonary vein, issuing from the descending thoracic aorta into the lower lung and the left hilar has abundant collateral vessels and the left lower lung hyperemia is obvious (Fig. 1b,1c). The whole operation time is 55 min and the amount of bleeding during the operation is about10ml. The thoracic drainage tube was removed 1 day after the operation, and the total drainage volume was about 50 ml and she was discharged 2 days after the operation. Reexamination of chest CT four month after discharge showed no abnormality(Fig. 1d). Comparison of preoperative and postoperative CT showed that the Isolated lung tissue were significantly smaller. On the postoperative follow-up of 4 months, these patient’s physical condition was excellent and free of any symptoms such as hemoptysis or cough.

**Discussion**

Pulmonary sequestration (PS) is a rare congenital respiratory malformation, also known as bronchopulmonary sequestration, accounting for 0.2–6.4% of Pulmonary developmental paraplasia[1, 3]. PS is usually classified as intralobar pulmonary sequestration(ILS) which shares the visceral pleura of an otherwise normal pulmonary lobe, and extralobar pulmonary sequestration(ELS),in which the extralobar sequestration is separated from normal lung tissue with an independent pleural covering[1]. PS usually occurs in the lower lobe of both lungs and is more common in the lower lobe of the left lung.

PS is anatomically characterized by the presence of aberrant feeding arteries and non-functional lung tissue and feeding arteries come mainly from the systemic circulation. The feeding arteries of PS originate most frequently from thoracic aorta, followed by abdominal aorta, intercostal artery and subclavian artery and others [1, 3].

As the clinical symptoms of PS are not typical, misdiagnosis often occurs[1].PS appears symptomatic or asymptomatic manifestations. Common symptoms include fever, cough and other upper respiratory tract infection-related symptoms, and other lower respiratory tract infection-related symptoms, hemoptysis and other symptoms, and so on [1, 6]. The severity of symptoms is related to the initial site of feeding arteries, the location of nonfunctional lung tissue, the size of nonfunctional lung tissue, diameter of aberrant feeding artery, and whether there is a co-infection.

Imaging examination can clearly reveal aberrant feeding arteries, providing key information for preoperative diagnosis. DSA is the gold standard for the diagnosis of PS. Enhanced CT and CTA can clearly show aberrant feeding arteries, identify the initial site and shape of aberrant feeding arteries, and significantly reduce the risk of bleeding in the intraoperative blood vessels in the abnormal body circulation caused by insufficient preoperative evaluation[7, 8]. For suspected patients with PS, enhanced CT and CTA can effectively reduce misdiagnosis and missed diagnosis of PS. CTA was used in the diagnosis of PS in this case.

It is controversial that whether patients with PS should receive surgical intervention because data regarding the surgery outcome, long-term clinical course and quality of life are rare[8–11]. Patients with different symptoms of PS should receive different treatment strategies. It is generally accepted that
patients diagnosed with ILS whether asymptomatic or not and patients diagnosed with ELS with symptom should receive surgical intervention [10]. Non-operative management is better suited for asymptomatic ELS. Studies have shown that ELS is likely to regress spontaneously, supporting the safety of non-operative ELS management [4, 9, 11].

Surgical strategies for PS are challenging. Increasing numbers of surgical schemes have been proposed due to the disadvantages of traditional open surgery [12–15]. Studies have shown that thoracoscopic therapy for PS patients is feasible and safe and has advantages compared with traditional open surgery [13, 14]. Lobectomy is usually regarded as an effective surgical method for treating ILS. Considering the benign nature of PS, sublobectomy has its advantages of preserving more postoperative lung function compared with lobectomy. However, its long-term prognostic data are lacking [13, 14].

Then the surgical resection scope of ILS patients can be further reduced, there have been case reports of endovascular technique and hybrid surgery treatment for PS patients, but no large-scale studies have been conducted [12, 16]. Intravascular technology is less invasive, but carries a risk of endovascular complications and has no long-term prognosis. For PS patients with clinical manifestations were mainly non-infective symptoms, feeding artery mutilation may be preferable than endovascular techniques, especially when the aberrant feeding arteries are difficult to be treated with endovascular techniques.

This operation was performed by uniportal video-assisted thoracoscopy and compared with Lin et al.'s study [14], the aberrant feeding arteries were divided by uniportal video-assisted thoracoscopy has significantly shorter operation time, less intraoperative blood loss, fewer postoperative total drainage volume, and shorter postoperative thoracic drainage time. However, due to the isolated lung tissue was not removed there are risks of secondary infection in the isolated lung tissue and hemoptysis. There were no complications in the postoperative follow-up of the patient.

This study confirms that it is safe and effective to treat PS patients by the aberrant feeding arteries disconnection. PS can be treated by disconnecting aberrant feeding arteries to avoid further aggravation of the disease and thus avoid the removal of lung parenchyma. And we recommend that it is necessary to preoperative localization of abnormal blood vessels to avoid intraoperative hemorrhage caused by traumatized aberrant feeding arteries.

In the current study, we retrospectively analyzed the clinical data regarding surgical procedures, outcomes, and prognosis of this case of PS who was treated with aberrant feeding artery mutilation, with the aim to improve the awareness and an attempt at a new form of surgery.

**Conclusion**

Surgery is still the first choose for treatment of PS. When pulmonary lesion is small-sized, for PS patients with non-infectious symptoms, aberrant feeding artery mutilation can be attempted as an alternative to intravascular technology for the treatment of PS. Future large studies are required to further verify its long-term efficacy and scope of application.
Abbreviations

PS
pulmonary sequestration

ILS
intralobar pulmonary sequestration

ELS
extralobar pulmonary sequestration

Declarations

Ethics approval and consent to participate

This study was approved by the Ethics Committee of Second Xiangya Hospital of Central South University, Changsha, China.

Consent for publication

Written informed consent for publication of the clinical details and/or clinical images was provided by the patient.

Availability of data and materials

As a case report, all data generated or analysed are included in this article.

Competing interests

The authors have no conflicts of interest to declare in this work.

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Authors’ contributions

YKL drafted and edited this manuscript and analysed the patient data. WH edited this manuscript and analyzed the patient data. RYW and YH prepared and assessed the imaging images. PX and FLY analyzed the patient data WLL performed the surgery, edited this manuscript, and analysed the patient data.
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