Case Report

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Practical Considerations for Endovascular Treatment of Pulmonary Sequestration-Induced Hemoptysis: Two Case Reports and Review of Literature

폐분리증으로 인한 객혈의 혈관 내 치료를 위한 임상적 고찰: 2건의 증례 보고 및 문헌 검토

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Pulmonary sequestration is a rare condition in which a segment or lobe of dysplastic lung tissue develops without connecting to the airways and pulmonary arteries. Hemoptysis is a potentially fatal complication of pulmonary sequestration. The current report discusses two complex cases of pulmonary sequestration. The first involves a patient with coexisting partial anomalous pulmonary venous drainage, in which hemoptysis was effectively managed through embolization of the abnormal systemic artery. Embolization of the anomalous pulmonary vein was not performed because the left-to-right shunt was hemodynamically insignificant. The second case involved a patient with a multi-cystic lung mass and air-fluid levels. Although embolization of the abnormal systemic artery controls hemoptysis, it also leads to prolonged complications due to aggravated inflammation. When providing endovascular treatment for pulmonary sequestration, conducting a comprehensive evaluation using pretreatment imaging is necessary to tailor the approach to each patient's unique condition.

Index terms Hemoptysis; Therapeutic Embolization; Bronchopulmonary Sequestration; Scimitar Syndrome; Lung Abscess

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INTRODUCTION

Pulmonary sequestration is a rare condition in which a segment or lobe of dysplastic lung tissue develops without connecting to the airways and pulmonary arteries. Instead, it receives blood supply from systemic arteries (1). The sequestered lung tissue and the normal bronchial tree may form a connection that can cause recurrent lung infection or hemoptysis. Hemoptysis is a potentially fatal complication caused by high capillary pressure from systemic supplying arteries. While surgery is the traditional treatment for hemoptysis, emerging evidence suggests that endovascular embolization is an effective alternative (2). However, the uncommon nature of this condition makes it challenging to define standard practice for endovascular treatment. This report contributes to the ongoing discussion by describing two complex pulmonary sequestration cases requiring special attention.

CASE REPORTS

CASE 1

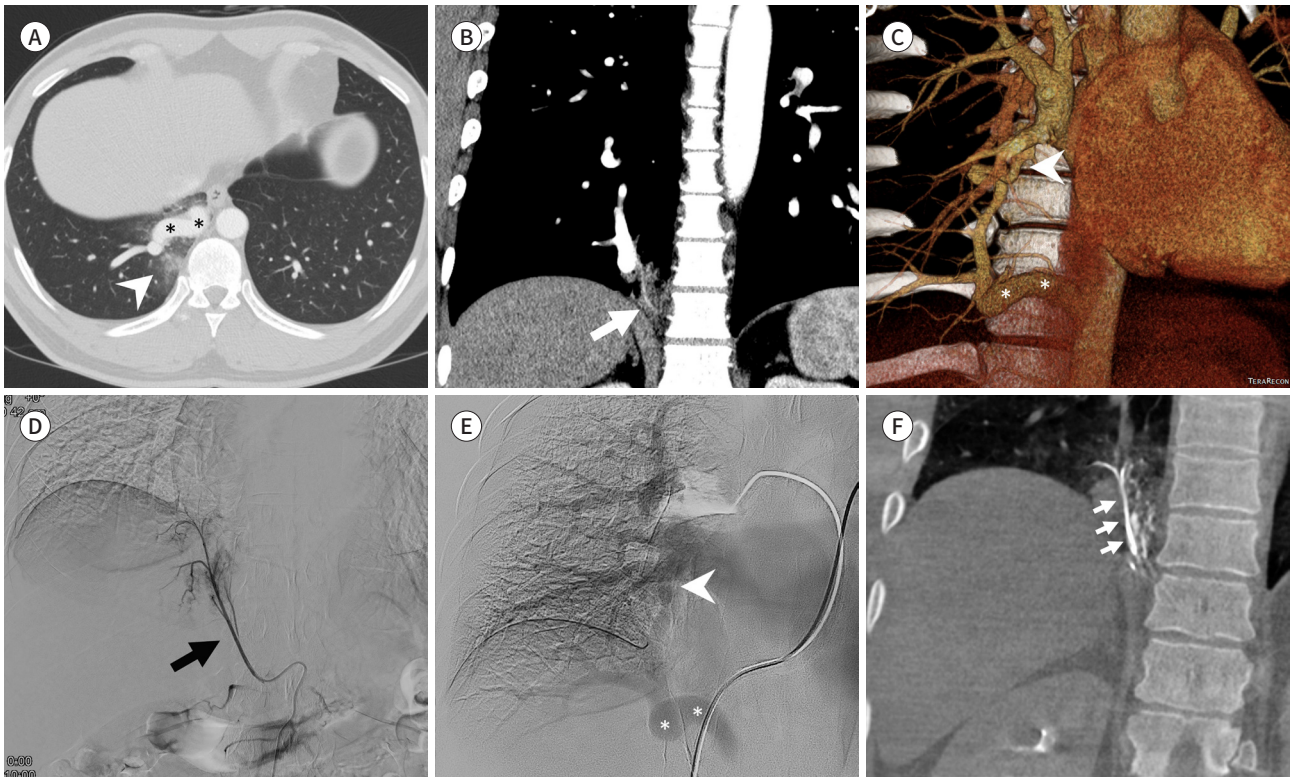
A 32-year-old male with no history of tuberculosis complained of hemoptysis and coughing for 2 days. Contrast-enhanced chest CT revealed an abnormal vascular structure extending from the descending thoracic aorta to the ground-glass attenuation of the right lower lung, indicating pulmonary sequestration (Fig. 1). There was also a connection between the right superior pulmonary vein and both the left atrium and inferior vena cava, suggesting partial anomalous pulmonary venous drainage (PAPVD). Bronchoscopy showed that the bleeding was localized to the posterior basal segment of the right lower lobe, and no definite endobronchial lesions were identified. Endoscopic instillation of cold saline and epinephrine failed to achieve hemostasis. The patient refused to undergo surgery and underwent endovascular treatment.

Under local anesthesia, the right common femoral artery and vein were accessed using 5-Fr vascular sheaths (Radifocus Introducer II, Terumo, Tokyo, Japan). Digital subtraction angiography of the abnormal systemic artery using a 5-Fr catheter (Mikaelsson catheter; Cook Medical, Bloomington, IN, USA) confirmed blood supply to an area of associated pulmonary sequestration. Selective embolization was performed using a 1.9-Fr microcatheter (Carnelian SI; Tokai Medical Products, Aichi, Japan) with an emulsion made of N-butyl cyanoacrylate (NBCA) and Lipiodol (Guerbet, Aulnay-sous-Bois, France) in a 1:3 ratio. During the delayed phase of right pulmonary angiography with a 5-Fr catheter (Davis catheter; Cook Medical), anomalous pulmonary veins were identified. Anomalous pulmonary veins were not embolized because the left-to-right shunt was not clinically significant (pulmonary-to-systemic flow ratio [Qp/Qs] < 1.5). Hemoptysis was immediately halted upon completion of the procedure, and the patient was discharged after 3 days.

CASE 2

A 33-year-old male was admitted to our hospital with chronic hemoptysis that lasted for 6 months. He had no other respiratory symptoms at the time of admission, but he reported a history of recurrent pneumonia since the age of 16. Chest radiography revealed multiple cys-

Fig. 1. Case findings of a 32-year-old male patient with pulmonary sequestration.
A, B. Contrast-enhanced chest CT scan shows tortuous anomalous pulmonary vein (asterisks) draining into the inferior vena cava and an abnormal systemic feeding artery (arrow) from descending thoracic aorta. Focal ground glass opacity (arrowhead) is also noted in the right lower lobe.
C. The volume-rendered image demonstrates dual venous drainage into the right superior pulmonary vein (arrowhead) and inferior vena cava (asterisks). No normal right lower pulmonary vein is visible.
D, E. Digital subtraction angiography confirms abnormal systemic artery (arrow) and anomalous pulmonary veins (arrowhead, asterisks).
F. Post-embolization CT confirms glue cast (arrowheads) in the abnormal systemic artery.

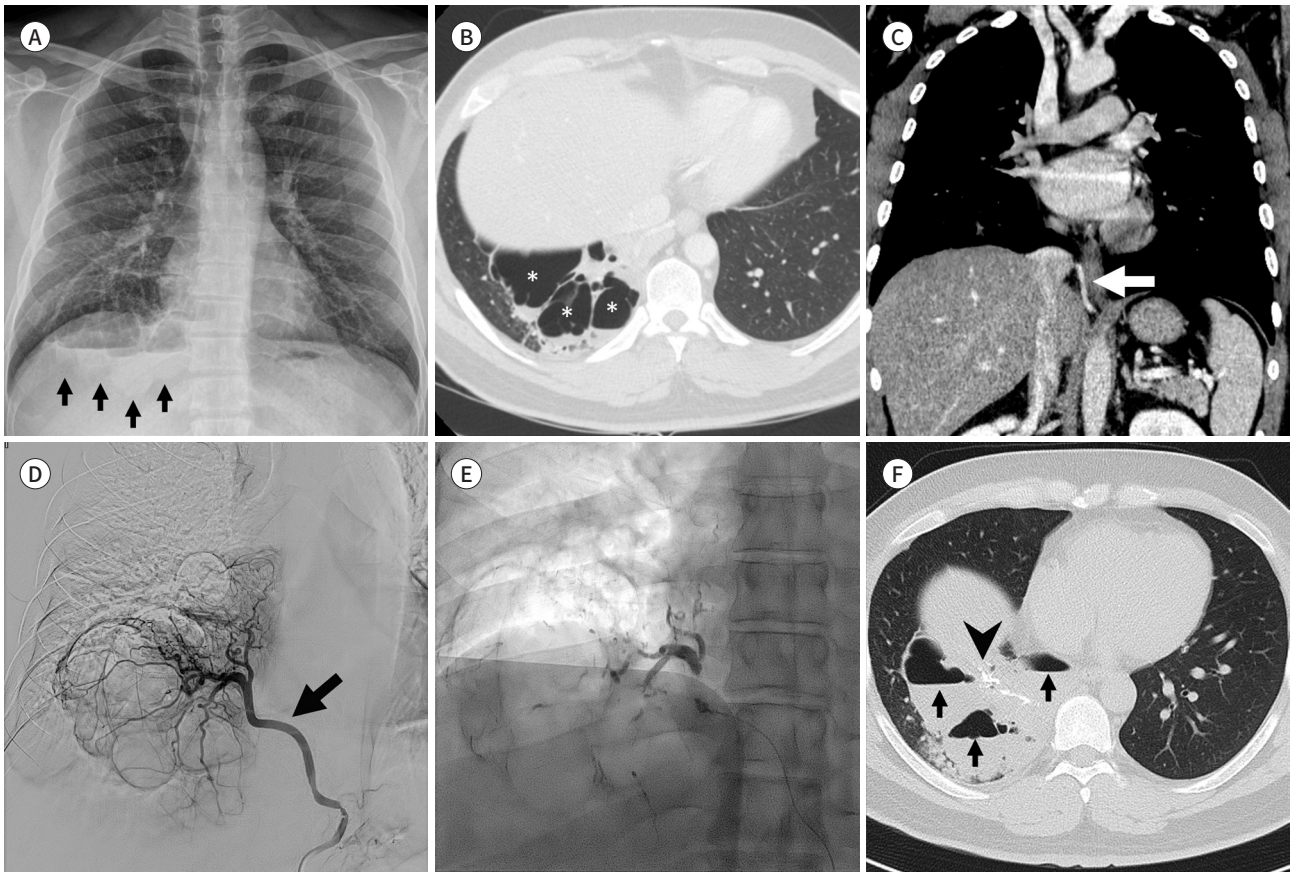


tic lesions with air-fluid levels in the lower right lung (Fig. 2). Contrast-enhanced chest CT revealed an abnormal vascular structure extending from the celiac axis to the multi-cystic mass, suggesting pulmonary sequestration. The multi-cystic mass contained air-fluid levels along with adjacent consolidations. Despite being informed of the risk of worsening inflammation following the procedure, the patient opted for endovascular treatment rather than surgery.

The right common femoral artery was accessed via a 5-Fr vascular sheath under local anesthesia. Digital subtraction angiography with a 5-Fr catheter (Rosch Hepatic; Cook Medical) demonstrated blood supply to a region of pulmonary sequestration through an abnormal systemic artery. Using an emulsion containing NBCA and Lipiodol in a 1:5 ratio, selective embolization was performed using a 1.9-Fr microcatheter. The patient experienced post-embolization fever for 9 days and was discharged with antibiotics. However, he returned 2 weeks later with recurrent fever. Follow-up chest CT revealed a marked increase in fluid in the multi-cystic lesion, suggesting infarcted sequestration. The patient was discharged 10 days after antibiotic treatment.

Our institution's Institutional Review Board (IRB No. 2023-11-018) approved this study and waived the requirement for informed consent.

Fig. 2. Case findings of a 33-year-old male patient with pulmonary sequestration.
A. Chest radiography reveals multiple cystic lesions with air-fluid levels (arrows) in the right lower lung.
B, C. Contrast-enhanced chest CT scan demonstrates thin septations with air-fluid levels (asterisks) in multi-cystic lesions. An abnormal vascular structure (arrow) from the celiac axis is also visible.
D. Digital subtraction angiography of the celiac axis confirms abnormal systemic artery (arrow) supplying multi-cystic lesions.
E. After embolization, a glue cast is noted in the abnormal systemic artery.
F. Follow-up chest CT scan reveals a marked increase of fluid (arrows) in the multi-cystic lesion. Glue cast (arrowhead) is also noted in the abnormal systemic artery.



DISCUSSION

Pulmonary sequestration represents 0.15%–6.4% of all congenital lung malformations (3). Patients with pulmonary sequestration may have no symptoms, and the diagnosis is often made incidentally. However, the intralobar sequestered lung tissue can communicate with the normal bronchial tree, which can cause recurrent lung infections or hemoptysis (4). Although rare, hemoptysis in these patients can range from intermittent to massive life-threatening events. The cause of hemoptysis is believed to be elevated systemic pressure in the feeding arteries, which may also be fragile because of medial hypertrophy and endarteritis.

The conventional treatment for symptomatic pulmonary sequestration in adults involves the surgical removal of abnormal tissue or lobectomy. Endovascular treatment has been sporadically reported to be a less invasive alternative to surgery (5, 6). This option is advantageous in situations such as ours, particularly for patients with comorbidities or those who refuse to

undergo surgery. The primary concern was hemoptysis, which was successfully managed in both cases. However, the unique aspects of our patients differ from those of previous cases, thus warranting further discussion.

Since pulmonary sequestration is often associated with other cardiovascular anomalies, imaging studies are vital in revealing coexisting conditions that might complicate treatment (7). The venous connection is particularly important, as PAPVD can affect not only the sequestered segment but also the entire ipsilateral lung, thereby making endovascular treatment far more complex. A pulmonary-to-systemic flow ratio >1.5:1 was considered significant and indicative of the need for surgical or endovascular correction (8). If a patient presents with clinical symptoms or a hemodynamically significant left-to-right shunt, simultaneous embolization of the abnormal systemic artery and anomalous pulmonary vein may be considered (9).

Based on the current literature, major long-term complications following endovascular treatment of pulmonary sequestration are rare (10). Our second case suggests that patient-specific factors, particularly underlying inflammation, may significantly affect the outcome. The air-fluid level is an important imaging indicator; however, it is not exclusively diagnostic of inflammation or infection. Therefore, this finding should be interpreted in the context of the patient's clinical history and symptoms.

Literature regarding embolic materials for pulmonary sequestration is limited, but polyvinyl alcohol particles, microcoils, and vascular plugs are commonly used (2, 5, 6). We used NBCA as an embolic agent because the goal was the complete cessation of hemoptysis, similar to bronchial artery embolization. As expected, NBCA effectively controlled hemoptysis; however, it appeared to contribute to the extensive infarction of the sequestration tissue. Microcoils or vascular plugs were not considered because they can prevent future procedures for recurrent hemoptysis due to proximal embolization.

In conclusion, endovascular treatment of pulmonary sequestration requires a thorough evaluation of pretreatment imaging findings to understand the unique details of each patient's condition and accordingly choose the most effective approach.

Author Contributions

Writing—original draft, K.M., L.H.N.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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폐분리증으로 인한 객혈의 혈관 내 치료를 위한 임상적 고찰: 2건의 증례 보고 및 문헌 검토

김민희 · 이형남* · 김승수 · 박찬호 · 박혜림

폐분리증은 이형성 폐 조직의 한 부분 또는 엽이 기도와 폐동맥에 연결되지 않고 발달하는 드문 질환이다. 객혈은 폐분리증의 잠재적으로 치명적인 합병증이다. 이번 증례 보고에서는 두 가지 복잡한 폐분리증 증례들을 기술하고자 한다. 첫 번째는 부분 폐정맥 환류 이상이 동반된 환자로, 객혈은 비정상적 체동맥의 색전술을 통해 효과적으로 치료되었다. 좌우 단락은 혈역학적으로 의미 있게 크지 않아서 비정상적인 폐정맥의 색전술은 시행되지 않았다. 두 번째 증례는 다낭성 폐 종괴 내에 공기액체층이 보였던 환자이다. 객혈은 비정상적 체동맥의 색전술로 조절할 수 있었지만, 시술 후 염증이 악화되어 합병증이 장기화되었다. 폐분리증의 혈관 내 치료를 고려할 때는, 각 환자의 고유한 특성에 맞는 치료 전략을 세우기 위해 치료 전 영상에 대한 포괄적인 평가가 필요하다.

순천향대학교 의과대학 천안병원 영상의학과