Case Report
pISSN 1738-2637
J Korean Soc Radiol 2012;67(2):105-108

대한영상의학회지

Endovascular Treatment of Intralobar Pulmonary Sequestration with Vascular Plug: A Case Report[1]

Vascular Plug를 이용한 내엽성 폐분리증의 동맥내 중재적 치료: 증례 보고[1]

Yun Jung Kim, MD[1], Young Kwon Cho, MD[1], Jae Hyung Lee, MD[2], Jeong Joo Woo, MD[1], Yun Sun Choi, MD[1]

Departments of [1]Radiology, [2]Internal Medicine, Eulji Hospital, Eulji University School of Medicine, Seoul, Korea

Intralobar pulmonary sequestration is a rare entity, which requires surgical resection as the treatment of choice. Recently, endovascular treatment for occlusion of the feeding artery has been frequently used with the increased availability regarding a variety of embolic materials, such as gelatin sponge particles, polyvinyl alcohol particles, and coils. Authors described the successful treatment of intralobar pulmonary sequestration with a Type II Amplatzer Vascular Plug and antibiotic-soaked gelatin sponge particles for complete occlusion of the feeding artery regarding intralolar pulmonary sequestration. Neither recanalization of the feeding artery for the sequestered lung nor other complications occurred during a 1-year follow-up.

Index terms
Pulmonary Sequestration
Endovascular Treatment
Intralobar

Received March 19, 2012; **Accepted** July 3, 2012
Corresponding author: Young Kwon Cho, MD
Department of Radiology, Eulji Hospital, Eulji University School of Medicine, 14 Hangeulbiseok-gil, Nowon-gu, Seoul 139-872, Korea.
Tel. 82-2-970-8290 Fax. 82-2-970-8346
E-mail: ykchoman@eulji.ac.kr

INTRODUCTION

Pulmonary sequestration is a congenital malformation involving a mass of non-functioning pulmonary tissue that does not communicate with the bronchial tree or the pulmonary arteries (1). Intralobar sequestration is commonly seen in young adults with no symptoms or nonspecific symptoms, such as chest pain, dyspnea, and wheezing. Recently, several case reports documented that endovascular embolization with coils is a safe and effective alternative treatment replacing surgery when dealing with pulmonary sequestration (2-4). Also, the use of new occluding devices has been associated with high success rate and efficacy for the embolization of the aberrant artery to the intralobar sequestration from the aorta (5, 6). There has been no established regimen for endovascular treatment until now. As far as we know, we are the first to describe the successfully treated case of intralobar pulmonary sequestration by occlusion of the feeding artery with type II Amplatzer Vascular Plugs and antibiotic-soaked gelatin sponge particles in an adult.

CASE REPORT

A 30-year-old woman complained of generalized myalgia, fever, and intermittent cough for 3 days with a history of recurrent pneumonia since childhood. She had a negative result for the influenza virus then she was treated with oral antibiotics for 3 days while hospitalized.

The chest X-ray revealed a large ill-defined consolidation in the left lower lung (Fig. 1A). Contrast-enhanced axial computed tomography images showed pulmonary sequestration represented as a heterogeneous mass containing multifocal cystic portions and a feeding artery, which has numerous parenchymal branches into the sequestered lung, originating from the lower thoracic aorta to the left lower lung on the three-dimensional recon-

structed volume rendered technique (Fig. 1B).

Selective arteriograhy with a 5 F diagnostic catheter (Terumo Corp., Tokyo, Japan) showed a diameter of 8.5 mm, feeding the artery to the lesion with multiple small irregular distal branches from the lower thoracic aorta to left lower lung field (Fig. 1C).

After selection of the feeding artery with an 8 F Envoy guiding catheter (Cordis, Johnson & Johnson, San Diego, CA, USA), we infused approximately 1000 EA gelatin sponge particles (approximately 1 mm^3, Spongostan; Johnson & Johnson Medical, Gargrave, UK) that had been soaked in 4 g of cephtriaxone (Yuhan Pharm. Co., Ltd., Seoul, Korea) into the distal branches of the feeding artery via a 2.8 F microcatheter (Terumo Corp., Tokyo, Japan) for the complete embolization of distal branches and to also decrease the risk of infection.

Since the proximal diameter of the feeding artery was very large for coil embolization and the high risk of distal migration and incomplete packing of coil materials, we chose the type II Amplatzer Vascular Plug as the embolic material.

Firstly, we deployed an 8 mm plug into the 5 mm sized distal trunk of the feeding artery after the infusion of a large amount of gelatin sponge particles to multiple distal branches of the main trunk for the prevention of recanalized collateral vessels

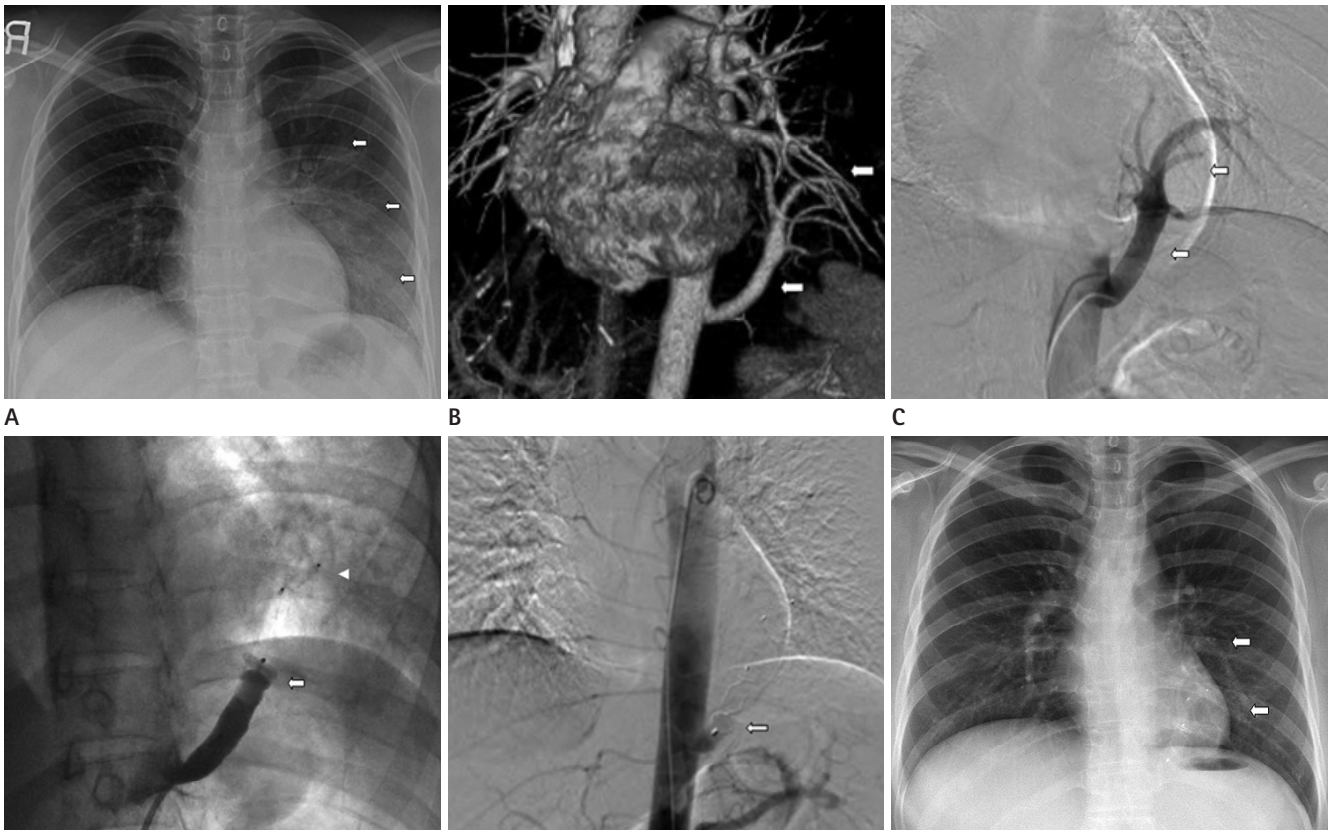

Fig. 1. Interventional embolization with type II Amplatzer Vascular Plug for the intralobar pulmonary sequestration in a 30-year-old woman.
A. Initial chest X-ray reveals large ill-defined consolidations in the left lower lung (arrows).
B. The three-dimensional reconstructed thoracic aortography with volume rendered technique shows a large feeding artery and numerous parenchymal branches of the sequestered lung, which originate from the lower thoracic aorta to the left lower lung field (arrows).
C. Selective arteriograhy of the feeding artery with a 5 F diagnostic catheter shows an 8.5 mm feeding artery (proximal) branching from the lower thoracic aorta to the left lower lung field (arrows) with drainage to the left atrium via the left inferior pulmonary vein. The shape of the branching vessels from the feeding artery has the appearance of a caput medusa with multiple smaller distal branches.
D. After infusion of a larger amount of gelatin sponge particles, which had been soaked in 4 g cephtriaxone, to the periphery of the distal branches of the feeding artery and loading of an 8 mm plug into the 5 mm sized distal lumen in the main trunk of the feeding artery (arrowhead), we deployed a 12 mm type II Amplatzer Vascular Plug into the 8 mm sized proximal lumen in the main trunk of the feeding artery (arrow).
E. Final thoracic aortography shows the totally occluded main feeding artery and the Vascular Plug placed within the proximal lumen of the feeding artery (arrow).
F. The 1-year follow-up chest PA shows the disappearance of the sequestered lung lesion in the left lower lung (arrows).

that might cause backflow, then we deployed a 12 mm type II Amplatzer Vascular Plug into the 8 mm sized proximal lumen in the main trunk of the feeding artery (Fig. 1D).

After 20 minutes following the procedure, the final thoracic aortography showed a total occlusion of the feeding artery by the Vascular Plugs in the proximal lumen of the feeding artery (Fig. 1E). One day after the procedure, cough and left lower chest pain were gradually resolved and the patient was discharged on the following day.

The 1-year follow-up chest PA demonstrated almost complete removal of soft tissue density of intralobar pulmonary sequestration in the left lower lung (Fig. 1F).

DISCUSSION

Intralobar pulmonary sequestrations receive systemic arterial supply from the thoracic or abdominal aorta or its branches. They share common visceral pleura with the parent lobe. The intralobar type of pulmonary sequestration is commonly seen in young adults, who often present with recurrent pneumonia as in our case, whereas the extralobar type is commonly seen in the pediatric group. Intralobar pulmonary sequestrations account for approximately 75% of cases and they are mostly located at the left lower lobe, while extralobar sequestrations commonly present at the posterior costodiaphragmatic sulcus between the left lower lobe and the left hemidiaphragm with other congenital anomalies (7, 8). The arterial supply to the sequestered lung most commonly arises from the descending thoracic aorta (73%) and the upper abdominal aorta (18.7%), and less frequently from the intercostal arteries, subclavian artery, internal thoracic artery, and rarely from the superior mesenteric artery (9, 10).

Surgery is the treatment of choice, namely sequesterectomy for the extralobar type and lower lobectomy for the intralobar type. If the patient is asymptomatic, surgical treatment is debatable in point of spontaneous regression of pulmonary sequestration.

Few case reports have shown equivalent results for successful endovascular embolization and surgery. Variable occluding materials have been used including gelatin sponge particles, polyvinyl alcohol particles, coils, and the Amplatzer Vascular Plug (2-6).

We performed endovascular occlusion of the large feeding artery using the type II Amplatzer Vascular Plug, which induced more rapid occlusion than the old Amplatzer Vascular Plug due to its unique multi-segmented and multi-layered design for more effective occlusion of vessels. We used an additional smaller type II Amplatzer Vascular Plug for more successful embolization of the main trunk regarding the feeding artery. In this setting, incomplete embolization and non-regression of sequestered tissue frequently occurred in 25-47% of cases, requiring additional surgery (11). So we also used antibiotic-soaked gelatin sponge particles (with a volume of approximately 1 mm^3 each) for occlusion of the smaller distal branches because of the presenting signs and symptoms of lung inflammation, the risk of infarction and the consequent abscess formation of the embolized sequestered lung (3, 12).

Until now, there has been scarce literature on favorable late outcomes following endovascular embolization for pulmonary sequestration with coils and gelatin sponge particles and no report on the long-term outcomes when using the Amplatzer Vascular Plug (3, 11). Hwang et al. (5) reported the successful occlusion of the aberrant artery to the intralobar sequestration with an old Amplatzer Vascular Plug, which had been used for the occlusion of a variety of pulmonary arteriovenous fistulas, coronary fistulas, PDA, portal vein and internal iliac artery aneurysms.

In conclusion, the endovascular embolization using the type II Amplatzer Vascular Plugs combined with the infusion of antibiotic-soaked gelatin sponge particles into the parenchymal branches of the feeding artery for pulmonary sequestration seems to be a safe and suitable alternative treatment for the intralobar pulmonary sequestration in adults.

REFERENCES

1. DeParedes CG, Pierce WS, Johnson DG, Waldhausen JA. Pulmonary sequestration in infants and children: a 20-year experience and review of the literature. *J Pediatr Surg* 1970;5:136-147
2. Madhusudhan KS, Das CJ, Dutta R, Kumar A, Bhalla AS. Endovascular embolization of pulmonary sequestration in an adult. *J Vasc Interv Radiol* 2009;20:1640-1642
3. Lee KH, Sung KB, Yoon HK, Ko GY, Yoon CH, Goo HW, et al.

Transcatheter arterial embolization of pulmonary sequestration in neonates: long-term follow-up results. *J Vasc Interv Radiol* 2003;14:363-367
4. Tokel K, Boyvat F, Varan B. Coil embolization of pulmonary sequestration in two infants: a safe alternative to surgery. *AJR Am J Roentgenol* 2000;175:993-995
5. Hwang HK, Tsai YS, Lin SM, Chen MR. Occlusion of an aberrant artery to an intralobar pulmonary sequestration using an Amplatzer Vascular Plug. *Pediatr Pulmonol* 2008;43:933-935
6. Crushell E, Saidi A, al-Hassan A, Walsh K. Occlusion of an aberrant artery to a pulmonary sequestration using a duct occluder. *J Interv Cardiol* 2002;15:415-416
7. Abbey P, Das CJ, Pangtey GS, Seith A, Dutta R, Kumar A. Imaging in bronchopulmonary sequestration. *J Med Imaging Radiat Oncol* 2009;53:22-31
8. Bolca N, Topal U, Bayram S. Bronchopulmonary sequestration: radiologic findings. *Eur J Radiol* 2004;52:185-191
9. Savic B, Birtel FJ, Tholen W, Funke HD, Knoche R. Lung sequestration: report of seven cases and review of 540 published cases. *Thorax* 1979;34:96-101
10. Hayasaka K, Saitoh T, Tanaka Y. Intralobar pulmonary sequestration receiving arterial supply from the superior mesenteric artery: a case report. *Comput Med Imaging Graph* 2006;30:135-137
11. Chien KJ, Huang TC, Lin CC, Lee CL, Hsieh KS, Weng KP. Early and late outcomes of coil embolization of pulmonary sequestration in children. *Circ J* 2009;73:938-942
12. Madoff DC, Denys A, Wallace MJ, Murthy R, Gupta S, Pillsbury EP, et al. Splenic arterial interventions: anatomy, indications, technical considerations, and potential complications. *Radiographics* 2005;25 Suppl 1:S191-S211

Vascular Plug를 이용한 내엽성 폐분리증의 동맥내 중재적 치료: 증례 보고[1]

김윤정[1] · 조영권[1] · 이재형[2] · 우정주[1] · 최윤선[1]

내엽성 폐분리증은 매우 드문 질환으로 주로 수술적 치료를 시행하였으나 최근 젤라틴 스펀지, polyvinyl alcohol 및 코일 등의 다양한 색전물질의 사용이 가능해지면서 급양동맥을 폐쇄시키는 동맥내 중재적 치료가 자주 이용되고 있다. 저자들은 Type II Amplatzer Vascular Plug와 항생제를 흡수시킨 젤라틴 스펀지를 이용하여 내엽성 폐분리증의 급양동맥을 성공적으로 폐쇄시켜서 치료했던 환자에 대해 기술하였다.

을지대학교 의과대학 을지병원 [1]영상의학과, [2]내과