Transcatheter arterial embolization of anomalous systemic arterial supply to the basal segment of the lung

Fumie Sugihara¹, Satoru Murata¹, Fumio Uchiyama², Jun Watari², Eiko Tanaka², Natsuka Muraishi², Etsuko Satoh³ and Shin-ichiro Kumita¹

Abstract
We describe the case of a 67-year-old woman with an anomalous systemic arterial supply to the basal segment of the lung, which was managed successfully by transcatheter arterial embolization (TAE) with microcoils. Her chest computed tomography (CT) scan showed diffuse ground-glass opacity in the left lower lobe, no bronchial abnormalities, and blood supply from an anomalous artery originating from the descending thoracic aorta, with drainage to the normal pulmonary vein. We successfully performed TAE under balloon occlusion of the anomalous artery, without complications. TAE is a minimally invasive, safe, and valuable method, and could be used as first-line treatment in such cases.

Keywords
Lung, thorax, interventional, embolization, congenital

Introduction
Anomalous systemic arterial supply to the basal segment of the lung is a rare congenital vascular malformation (1). An aberrant artery arising from the descending thoracic aorta supplies the normal basal segment of the lung without sequestration. Common clinical symptoms include hemoptysis, exertional dyspnea, chest pain, recurrent infection, and congestive heart failure due to pulmonary hyperperfusion or hypertension. Surgery has been the traditional treatment for this disorder, but recently transcatheter arterial embolization (TAE) has been attempted as a minimally invasive therapy. Here we describe a case with an anomalous systemic arterial supply to the basal segment of the lung that was successfully treated with coil embolization.

Case report
A 67-year-old woman had an abnormal shadow on her chest computed tomography (CT) scan that was noticed by her health check service. She had recently experienced mild fatigue, but reported no previous illness and had no hemoptysis, hemoptysis, or dyspnea. She was a non-smoker. The physical examination revealed no abnormalities in either breath or heart sounds. Blood examination, including arterial blood gas measurement, was normal; electrocardiographic and pulmonary function test findings were within the normal range.

A chest radiograph showed a band-like opacity in the lower lung field. Subsequently, contrast-enhanced CT showed diffuse ground-glass opacity (GGO) and thickened interlobular pleura in the left lower lobe, indicating pulmonary congestion (Fig. 1a). The normal pulmonary artery that accompanies a bronchus could not be identified in the left basal segment of the lung, which was supplied by an anomalous artery.

¹Department of Radiology, Nippon Medical School, IVR Center, Tokyo, Japan
²Department of Radiology, Ebina General Hospital, Kanagawa, Japan
³Nippon Medical School Graduate School Division of Pulmonary Medicine, Infection Diseases and Oncology, Tokyo, Japan

Corresponding author:
Fumie Sugihara, Department of Radiology, Nippon Medical School, IVR Center, 1-1-3 Sendagi, Bunkyo-ku, Tokyo 113-8602, Japan.
Email: giiorcubgogo@nms.ac.jp
7 mm in diameter, originating from the descending thoracic aorta (Fig. 1b). The pulmonary bronchi and veins were anatomically normal. An echocardiographic examination showed no dilatation of the right atrium or right ventricle, nor was any enlargement of the pulmonary artery observed. Echocardiography showed a tricuspid regurgitation pressure gradient (TRPG) of 32 mmHg, and a mean pulmonary artery pressure of 42.4 mmHg (normal range, 9–18 mmHg). These values did not exclude right heart strain. On lung perfusion scintigraphy, a perfusion defect was observed in the lower lobe of the left lung. The patchy perfusion suggesting pulmonary hypertension was not seen. Moreover, the shunt index of the lung perfusion scintigraphy was 10.1% (normal, <15%). Lung ventilation scintigraphy showed no radiotracer defects. An interventional procedure was performed under local anesthesia. The right common femoral artery and right femoral vein were punctured, and 4- and 5-French sheaths (Super Sheath; Medikit, Tokyo, Japan) were inserted, respectively. A 4-French pigtail catheter (PTA catheter; Medikit, Tokyo, Japan) was advanced to the descending thoracic artery, and a 5-French pigtail catheter (PA-1 catheter; Medikit, Tokyo, Japan) was advanced to the pulmonary artery. Then, thoracic aortography and pulmonary arteriography were performed. The angiographic examinations confirmed the absence of pulmonary branches in the left basal segments (Fig. 2a), and showed an anomalous artery originating from the lower thoracic descending aorta (Fig. 2b), with the normal lower pulmonary vein flowing into the left atrium in the venous phase. The diameter of the inferior and upper pulmonary veins was almost equivalent, and no enlargement was apparent. Bronchial arteriography showed no anomalous artery. Thus, we confirmed the diagnosis of an anomalous systemic arterial supply to the basal segment of the lung.

The patient was informed of the treatment options, including observation, surgery, and TAE, by thoracic surgeons, respiratory physicians, and interventional radiologists. Despite the lack of long-term results and the need for follow-up, she chose treatment with TAE because it is a minimally invasive procedure.

Under local anesthesia, the right common femoral artery was punctured to insert a 5-French sheath (Super Sheath; Medikit, Tokyo, Japan). A 5-French balloon catheter, 9 mm in diameter (Selecon MP Catheter II; Terumo Clinical Supply, Gifu, Japan) was advanced to the origin of the anomalous artery in order to avoid migration of the coils from the main trunk. A microcatheter (Renegade; Boston Scientific, Natick, MA, USA) was then advanced to the distal section of the anomalous artery. Under balloon occlusion (Fig. 2c), we performed embolization from the distal site of the main trunk to the origin, using five fibered IDC coils (Interlock; Boston Scientific, Natick, MA, USA) and 13 platinum coils (Tornade; Cook, Bloomington, IN, USA). Post-embolization angiography showed disappearance of the anomalous artery (Fig. 2d). After the embolization, the patient reported no chest pain or discomfort and she was discharged 2 days after the procedure. Her postprocedure course was good and her fatigue diminished. A follow-up CT after 9 months showed that the GGO of the lung field in the left basal segment had not disappeared, but was slightly decreased. Echocardiography revealed that the TRPG had decreased from 32 mmHg to 22 mmHg, and mean pulmonary artery pressure from 42.4 mmHg to 27 mmHg. Other echo parameters were almost the same as before TAE.

Discussion
An anomalous systemic arterial supply to the basal segment of the lung has been classified with intralobar...
pulmonary sequestration as Pryce's type I since 1946 (2). This anomaly, as a congenital vascular malformation, is now considered to be an independent disease from pulmonary sequestration. It is characterized by an anomalous systemic artery, drainage to the normal pulmonary vein, absence of the normal pulmonary artery, and no anomaly of the bronchus. The basal segments receive their systemic arterial blood supply from the aorta, which increases pulmonary blood pressure. In most cases, this congenital abnormality appears in the left basal segment of the lung, and the anomalous artery is usually a single vessel arising from the left side of the descending thoracic aorta. Very rarely, the congenital abnormality is in the right basal segment of the lung (3,4), and is associated with two or more anomalous arteries that arises from the proximal abdominal aorta or the celiac trunk (3). As a result of improvements in diagnostic imaging technology, the existence of the anomalous artery, the absence of a normal pulmonary artery and a normal branch of the bronchus can all be demonstrated using only a chest CT. Angiography is also a very useful examination, which can evaluate the hemodynamic status and the presence or absence of a shunt. In addition, lung ventilation and perfusion scintigraphy can evaluate the ventilation capability and the degree of pulmonary hypertension, as well as revealing small shunts that are not detected by CT or angiography.

The main complaints reported by patients with this congenital abnormality were hemosputum and hemoptysis. Some patients exhibit no warning symptoms and are identified at an advanced age because of an unusual radiological chest shadow, as in our case (5). This abnormal vessel was supplied by a systemic artery and drained to the pulmonary vein; as it created a left-left shunt, it was likely to cause pulmonary hypertension, extensive hemoptysis, and heart failure in the future. Moreover, since there has been a report of extensive hemoptysis causing death while the patient was under observation (6), early treatment would appear to be preferable. In our case, endovascular treatment was necessary in order to prevent the progression of pulmonary hypertension and the possibility of sudden death from extensive hemoptysis.

The main treatment for this abnormality has been surgical excision, such as lung lobectomy, ligation, or division of the anomalous artery. However, in recent years, an increasing number of cases have been treated

Fig. 2. Angiographic findings and treatment. (a) Pulmonary arteriography shows the absence of pulmonary branches in the left basal segments. (b) Thoracic aortography shows an anomalous artery arising from the lower thoracic descending aorta. (c) Selective angiography of the anomalous artery under balloon occlusion (arrow) shows irregularities in the peripheral branches. (d) Thoracic aortography post embolization shows disappearance of the anomalous artery.
using embolization (4,7,8). The standard embolization materials are microcoils, and several cases using an Amplatzer vascular plug have been reported (4). Migration is considered to be a major complication in the use of these embolization materials (9). In our case, since there was a risk of reflux to the thoracic or abdominal aorta, the procedure required careful judgment and skilled technique. By controlling the blood flow using a balloon catheter, a safe coil embolization procedure could be performed without migration of the coils. Liquid embolization materials are not recommended because of the high risk of extensive embolization, possibly leading to pulmonary embolization and pulmonary infarction.

According to the recent literature, chest pain and discomfort are often reported as complications after TAE (7,8). These symptoms are considered to be consequences of ischemic lung tissue after embolization. In our case, no complications occurred after TAE. Usually, the embolized area receives collateral flow from the pulmonary artery or bronchial arteries, so that any symptoms are reduced with the development of collateral pathways (10). However, since there have been few embolization cases that have undergone long-term observation, it is unknown whether long-term complications appear or not. In our case, there were no complications by 12-month follow-up.

In conclusion, TAE as a treatment of an anomalous systemic arterial supply to the basal segment of the lung is a minimally invasive, safe, and valuable method, without serious complications. Therefore, TAE might be considered as a first-line treatment in such cases, as an alternative to surgery.

References
1. Painter RL, Billig DM, Epstein I. Anomalous systemic arterialization of the lung without sequestration. N Engl J Med 1968;279:866–867.
2. Pryce DM. Lower accessory pulmonary artery with intralobar sequestration of lung: a report of seven cases. J Pathol 1946;58:457–467.
3. Akiba T, Marushima H, Kamiya N, et al. Anomalous systemic arterial supply to the basal segments of the lung with three aberrant arteries. Gen Thorac Cardiovasc Surg 2012;60:118–121.
4. Jiang S, Shi JY, Zhu XH, et al. Endovascular embolization of the complete type of anomalous systemic arterial supply to normal basal lung segments: a report of four cases and literature review. Chest 2011;139:1506–1513.
5. Esaki T, Matsumoto M, Sugahara K, et al. An elderly case of anomalous systemic arterial supply to the normal basal segment of the left lower lobe and summary of reports of the disease in Japan. Nihon Kokyuki Gakkai Zasshi 2011;49:528–533.
6. Rubin EM, Garcia H, Horowitz MD, et al. Fetal massive hemoptysis secondary to intralobar sequestration. Chest 1994;106:954–955.
7. Saida T, Ninomiya H, Hojo F, et al. Systemic arterial supply to the normal basal segments of the left lower lobe treated by coil embolization, with long-term follow-up. Radiat Med 2006;24:365–368.
8. Anil G, Taneja M, Tan AG. Endovascular treatment of isolated systemic arterial supply to normal lung with coil and glue embolisation. Br J Radiol 2012;85:83–86.
9. Etezadi V, Gandhi RT, Benenati JF, et al. Endovascular treatment of visceral and renal artery aneurysms. J Vasc Interv Radiol 2011;22:1246–1253.
10. Guo YM, Zhu L, Yang J, et al. An experimental study on the repair mechanism after pulmonary embolism. Zhonghua Jie He He Hu Xi Za Zhi 2004;27:723–726.