Transarterial Embolization for Life-Threatening Spontaneous Hemopneumothorax

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Abstract
Two patients with life-threatening spontaneous hemopneumothorax had developed transpleural, nonbronchial, systemic collaterals from branches of the subclavian artery. It was thought that the collaterals had ruptured due to a collapsed lung and pneumothorax. Transarterial embolization of these arteries was performed, and hemostasis was obtained with no complications. A 37-year-old man underwent embolization of branches of the left internal thoracic artery and the left ascending cervical artery using gelatin sponges and metallic coils. A 33-year-old man underwent artery embolization for extravasation from a branch of the right thyrocervical trunk using n-butyl cyanoacrylate mixed with Lipiodol. Transarterial embolization of a transpleural, nonbronchial, systemic artery provided effective hemostasis in life-threatening spontaneous hemopneumothorax.

Key words: Spontaneous hemopneumothorax, Pneumothorax, Embolization, Bronchial artery

Introduction
Massive bleeding is sometimes observed in patients with spontaneous pneumothorax, known as “spontaneous hemopneumothorax.” Spontaneous hemopneumothorax is reported to occur in 3.7% of cases of spontaneous pneumothorax [1]. Typically, video-assisted thoracic surgery is used to identify the source of bleeding and to treat spontaneous hemopneumothorax. Although only 2 reported cases have been diagnosed with angiography, followed by embolization, it was thought that rapid hemostasis can also be achieved in such cases [1-4]. Transarterial embolization is a common procedure for life-threatening hemorrhage, and may also be a good option for hemostasis in spontaneous hemopneumothorax. There are, however, few reports of the angiographic findings and technique of transarterial embolization for spontaneous hemopneumothorax. Transarterial embolization for hemostasis and the angiographic findings of 2 cases of life-threatening spontaneous hemopneumothorax are reported.

Case 1
A 37-year-old man was transported to the emergency room with a chief complaint of respiratory discomfort. He then went into cardiopulmonary arrest. The chest radiograph on arrival showed bilateral pneumothorax. Bilateral chest tubes were inserted. A large amount of blood was drained via the left chest tube. Non-contrast-enhanced computed tomography (CT) showed bullae, and a bilateral spontaneous pneumothorax (Fig. 1a) was diagnosed. The hemoglobin level decreased from 10.4 mg/dL to 8.0 mg/dL in 3 hours, and hemorrhagic fluid (2,290 mL) was drained by the left chest tube, leading to a diagnosis of spontaneous hemopneumothorax. Transarterial embolization was performed to achieve hemostasis. All procedures were performed using an interventional radiology-CT angiography system (AXIOM...
A 37-year-old man visited the emergency outpatient department with a chief complaint of respiratory discomfort. Chest radiographs and chest CT demonstrated bullae and spontaneous pneumothorax. His hemoglobin level decreased from 13.1 mg/dL to 6.4 mg/dL in 3 hours, and his blood pressure decreased to 74/48 mmHg. Hemorrhagic fluid drainage (2,500 mL) was drained by the chest tube. Contrast-enhanced chest CT demonstrated hematoma and extravasation of contrast agent at the right lung apex. Transarterial embolization was performed to achieve hemostasis. All procedures were performed using an interventional radiology-CT angiography system (AXIOM). A 5-Fr, 25-cm sheath introducer (Super Sheath, Medikit) was inserted into the right femoral artery. A 5-Fr, headhunter-shaped catheter (Cook Medical) was used to select the subclavian artery, and a 2.2-Fr, microcatheter (SIRABE, Piolax Medical Devices, Yokohama, Japan) was inserted into the right femoral artery. A 5-Fr, 25-cm sheath introducer (Super Sheath, Medikit) was inserted into the right femoral artery. A 5-Fr, headhunter-shaped catheter (Cook Medical) was used to select the subclavian artery, and a 2.2-Fr, microcatheter (SIRABE, Piolax Medical Devices, Yokohama, Japan) was inserted to select the left internal thoracic artery and left ascending cervical artery.

Angiography demonstrated numerous dilated and vine-like abnormal blood vessels branching from the left internal thoracic artery and left ascending cervical artery (Fig. 1b, 1c). Contrast agent flowed into the pulmonary artery from these newly-developed blood vessels (Fig. 1d, 1e). Although contrast agent extravasation was not observed, the presence of blood vessels that continued from the chest wall into the lung was confirmed, and it was therefore determined that the rupture of these abnormal blood vessels due to the collapsed lung was the cause of bleeding. Embolization of the left ascending cervical artery and left internal thoracic artery was performed with 3 × 3-mm and 3 × 25-mm pieces of gelatin sponge (Serescue, Nippon Kayaku Co, Ltd, Tokyo, Japan) and 0.018-inch pushable coils (4-6 mm) (VortX Diamond-18, Boston Scientific, Marlborough, MA, USA). On aortography and subclavian arteriography, no other abnormal blood vessels and no extravasation were confirmed. Hemorrhagic fluid drainage from the chest tube stopped, and the hemoglobin level stabilized after embolization. No complications and no recurrence of hemopneumothorax were observed during 3 years of follow-up.

Case 2

A 33-year-old man visited the emergency outpatient department with a chief complaint of respiratory discomfort. Chest radiographs and chest CT (Fig. 2a) demonstrated bullae and spontaneous pneumothorax was diagnosed. His hemoglobin level decreased from 13.1 mg/dL to 6.4 mg/dL in 3 hours, and his blood pressure decreased to 74/48 mmHg. Hemorrhagic fluid (2,500 mL) was drained by the chest tube. Contrast-enhanced chest CT demonstrated hematoma and extravasation of contrast agent at the right lung apex (Fig. 2b). Transarterial embolization was performed to achieve hemostasis. All procedures were performed using an interventional radiology-CT angiography system (AXIOM). A 5-Fr, 25-cm sheath introducer (Super Sheath, Medikit) was inserted into the right femoral artery. A 5-Fr, headhunter-shaped catheter (Cook Medical, Bloomington, IN, USA) was inserted into the right femoral artery. A 5-Fr, headhunter-shaped catheter (Cook Medical) was used to select the subclavian artery, and a 2.2-Fr, microcatheter (SIRABE, Piolax Medical Devices, Yokohama, Japan) was inserted to select the left internal thoracic artery and left ascending cervical artery.

Contrast agent extravasation was not observed, the presence of new blood vessels (white arrow) is observed on left internal thoracic artery angiography. Embolization of the rupture of these abnormal blood vessels due to the collapsed lung was the cause of bleeding. Embolization of the left ascending cervical artery and left internal thoracic artery.

Figure 1. A 37-year-old man with a chief complaint of respiratory discomfort was transported to the emergency room. He was diagnosed with left spontaneous hemopneumothorax. a) Chest CT on arrival. Bilateral pneumothorax with bullae is seen. b) Digital subtraction angiography of the left internal thoracic artery. c) Digital subtraction angiography of the left ascending cervical artery. d) Digital subtraction angiography, early phase. Dilated and vine-like growth of abnormal blood vessels (white arrow) is observed on left internal thoracic artery angiography. e) Digital subtraction angiography, delayed phase. Contrast agent is seen flowing into the pulmonary artery (white arrowhead) from an abnormal vessel that developed from the left internal thoracic artery.

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A video-assisted thoracic surgery study reported that disjunction of transpleural, nonbronchial, systemic collateral arteries is the cause of bleeding in spontaneous hemopneumothorax [2]. This appears to correspond to the findings observed on subclavian artery angiography in the present case. In addition to the present study, there have been 2 other reported cases of spontaneous hemopneumothorax in which the causative blood vessel was identified by angiography, with observation of transpleural, nonbronchial, systemic collateral arteries [3, 5].

We carefully assessed the possibility of a feeder to the spinal cord on angiography prior to embolization, and used larger embolic materials (3 × 3-mm and 3 × 25-mm pieces of gelatin sponge and metallic coils in case 1) to prevent spinal infarction by distal embolization. However, it was thought that the risk of spinal infarction was not completely avoided because not all arteries feeding the spinal cord were detected by angiography. Complications of spinal cord infarction should always be considered with embolization in this area.

In case 2, it was possible to select the distal branch of the right thyrocervical trunk, and extravasation of contrast medium was confirmed. Therefore, n-butyl cyanoacrylate was used as an embolic material to reach the bleeding point. In both cases, there were no adverse events or complications of transarterial embolization. Video-assisted thoracic surgery has been commonly used for spontaneous hemopneumothorax. In emergency cases of spontaneous hemopneumothorax, however, we have considered transarterial embolization as a good option for hemostasis, similar to its use for other types of life-threatening hemorrhage. In addition, we have considered a branch of the subclavian artery as the most important source of bleeding in spontaneous hemopneumothorax. Thus, it was necessary to access the subclavian artery to obtain hemostasis in the present cases. To obtain hemostasis promptly and safely, selectively accessing the subclavian ar-
tery including the distal branch should be considered.

Transpleural, nonbronchial, systemic collateral arteries are commonly observed during bronchial artery embolization to treat hemoptysis in cavitary aspergillosis, cystic fibrosis, tuberculosis, and carcinoma, and advanced disorders of lung parenchyma are observed in all of these conditions [6]. Although bullae were observed in both of the present cases, no advanced lung parenchymal disorder was observed. Nonetheless, advanced growth of transpleural, nonbronchial, systemic collateral arteries was observed, and we postulated a specific cause for this phenomenon. It has been reported that patients who develop bullae have experienced ischemia at the lung apex due to rapid growth of the lung parenchyma during adolescence [7, 8]. We suspect that transpleural, nonbronchial, systemic collateral arteries develop to compensate for this ischemia. In addition, lung specimens resected from patients with spontaneous pneumothorax very often show vasculopathies, specifically pulmonary artery intimal fibrosis and pulmonary vein intimal fibrosis, at frequencies of 90% and 80%, respectively. These are thought to be caused by chronic inflammatory changes [9]. We postulated that transpleural, nonbronchial, systemic collateral arteries developed due to these changes, resulting in increased blood flow to the pulmonary artery due to the pressure gradient, resulting in the angiographic images observed in Case 1. Furthermore, we postulated that disjunction of the newly-developed transpleural, nonbronchial, systemic collateral arteries occurred when the lung separated from the chest wall due to the collapsed lung, causing massive bleeding.

**Conclusion**

The development of transpleural, nonbronchial, systemic collateral arteries at the subclavian artery branch was the cause of bleeding in 2 cases of spontaneous hemopneumothorax, and transarterial embolization provided effective hemostasis.

**Conflict of interest:** No authors have a financial interest to disclose.

No authors have a conflict of interest.

The study protocols for this retrospective analysis were approved by our institutional review board.

**References**