Hemoptysis Due to Type II Endoleak after Thoracic Endovascular Aortic Repair: Successful Treatment with Percutaneous CT-Guided Embolization

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Abstract
An 81-year-old woman presented with massive hemoptysis. She had a history of total arch replacement with an elephant trunk followed by concomitant antegrade thoracic endovascular aortic repair for the aortic arch and the descending aortic aneurysm. Computed tomography (CT) showed expansion of the aortic aneurysm with type II endoleak, lung parenchymal consolidation, and ground-glass opacity. An aortopulmonary fistula was suspected. Surgery posed a very high risk for the patient; hence, a less invasive approach was considered. Left subclavian arteriography revealed a type II endoleak. A transarterial approach would be difficult due to the small and tortuous access route and longer procedure time. Therefore, CT-guided puncture embolization was performed. She had no recurrence of hemoptysis for 1.4 years after the embolization.

Key words: Hemoptysis, CT-guided embolization, Type II endoleak, Thoracic endovascular aortic repair

1. Introduction
Primary management of a type II endoleak is close observation through imaging. However, type II endoleak embolization may be indicated in cases with an expanding [1] or symptomatic [2] aneurysm. Hemoptysis caused by a type II endoleak after thoracic endovascular aortic repair (TEVAR) is very rare [2]. In general, a type II endoleak is treated through transarterial or direct percutaneous puncture. Collateral circulation is not as well developed in the thoracic aorta as in the abdominal aorta [3]. Consequently, a transarterial approach after TEVAR is sometimes more difficult and requires more procedure time than direct percutaneous puncture [4]. Here we present a case of hemoptysis caused by a type II endoleak after TEVAR, which was treated with percutaneous computed tomography (CT)-guided embolization. The institutional review board at the participating institution waived the need for approval for this research.

2. Case Report
An 81-year-old woman had undergone total arch replacement with an elephant trunk followed by concomitant antegrade TEVAR (Zenith TX2; Cook Japan, Tokyo, Japan) for the aortic arch and the descending aortic aneurysm (70 mm in diameter) at another hospital 4 years ago. The aneurysm had decreased in size (55 mm in diameter) after the TEVAR. However, she presented with a gradually worsening hemoptysis since 1 month and was admitted to our hospital with severe hemoptysis (300 mL). On arrival, the hemoptysis temporarily stopped. After 5 days, she presented with recurrent and massive hemoptysis (300 mL) with hypotension. Her initial blood pressure was 68/41 mm Hg. Laboratory examination showed hemoglobin level of 7.8 g/dL, lowered from 9.6 g/dL on the previous day. She required intu-
bation and received a total of six units of packed red blood cells. A contrast-enhanced CT was performed. The CT images demonstrated an aneurysm progression (62 mm in diameter) with type II endoleak (Figure 1A), lung parenchymal consolidation, and a ground glass opacity in the lower lobe dominance with left pleural effusion (Figure 1B). The CT images showed type II endoleak from the left second intercostal artery. Active bleeding was observed predominantly from the left upper lobe bronchus on bronchoscopy. Stent graft removal and aortic reconstruction were planned because aorto-pulmonary fistula was suspected. However, the patient had poor tolerance for surgical treatment and preferred a less invasive approach. Left subclavian arteriography revealed type II endoleak from the left second intercostal artery via the highest intercostal artery (Figure 2). Sacography revealed the endoleak nidus and the left second intercostal artery via the highest intercostal artery (Figure 3A). We performed embolization using a 1.0 mL mixture of N-butyl-2-cyanoacrylate (NBCA) (Histoacryl; B. Braun, Melsungen, Germany) and iodized oil (lipiodol; Guerbet, Tokyo, Japan) (NBCA:lipiodol, 1:1) because the microcatheter for coil embolization could not be advanced into the left second intercostal artery via the small needle. The NBCA cast filling in the endoleak nidus was confirmed on a postoperative CT image (Figure 3C). Hemothrosis disappeared after embolization, and there were no neurological complications. Recurrence of hemothrosis was not observed. A follow-up CT at 1.4 years after embolization revealed that the aneurysm had decreased in size (50 mm in diameter), and the NBCA cast had filled the endoleak nidus (Figure 4A); there was no recurrent endoleak (Figure 4B).
3. Discussion

Type II endoleaks after TEVAR have been reported to occur in 10% (7/69) [3] of the patients; these rates are similar to those observed after EVAR [5]. Type II endoleak embolization can be attempted in cases with expanding [1] or symptomatic [2] aneurysms. To our knowledge, only two studies have reported hemoptysis caused by endoleaks [2, 6]. Aortobronchial or aorto-pulmonary fistula is assumed the background etiology in such cases. Synowiec et al. reported hemoptysis as the first symptom of a type IA endoleak after TEVAR [6]. The endoleak was treated by thoracotomy. Sueyoshi et al. have reported transarterial embolization for hemoptysis caused by a type II endoleak after TEVAR [2]. However, in the present case, transarterial embolization was difficult to perform due to the small and tortuous access route. Therefore, percutaneous CT-guided embolization was performed. Some studies have reported that cone beam CT is a good option to guide the direct percutaneous puncture of type II endoleak embolization after TEVAR [7, 8]. CT guidance minimizes the risk of visceral and vascular inju-
eries. Furthermore, intravenous contrast enhancement was performed in the present patient for visualizing the small endoleak nidus.

We also used the coaxial technique to puncture the endoleak nidus. Katada et al. reported the usefulness of this technique for direct percutaneous endoleak embolization [7]. They have reported that using this technique makes it possible to perform the second embolization immediately if the first embolization was insufficient. In our case, the endoleak nidus was relatively small (8 mm in diameter). Therefore, we used this technique to perform the second embolization. For the complete elimination of a type II endoleak, Katada et al. performed the embolization of the endoleak nidus and the proximal feeding artery [7]. However, Yu et al. have recently reported no significant differences in the rates of residual endoleak or endoleak nidus decrease/stabilization whether endoleak nidus embolization was performed alone or combined with feeding artery embolization [9].

Embolic agents for the direct percutaneous puncture embolization for type II endoleaks after TEVAR include NBCA-lipiodol alone [7, 10] and NBCA-lipiodol with coils [8]. NBCA is more feasible than coils because of the two disadvantages of coil embolization: the risk of coil compaction [7] and the endoleak channel remaining patent after coil embolization [5]. Meanwhile, Mussa FF et al. reported two advantages of embolization using NBCA-lipiodol with coils. First, the use of coils to occlude the main feeding vessel avoids accidental migration of NBCA-lipiodol mixture to the non-target end vessels. Second, the use of NBCA-lipiodol mixture to cast the aneurysm isolates the endoleak nidus from systemic pressure and increases the success of endoleak treatment by reducing the frequency of late recanalization [8]. Although there are no reports of embolization without NBCA in percutaneous thoracic type II endoleak embolization, it has been hypothesized that embolization using NBCA might be effective, and coil embolization alone should be avoided. In this case, we used a high concentration of NBCA-lipiodol (50%) to prevent paraplegia due to migration to communication with the anterior spinal artery because we could not occlude the feeding artery using coils. In the previous reports of percutaneous thoracic type II endoleak embolization with NBCA-lipiodol alone, a high concentration of NBCA-lipiodol (44-50%) was used [7, 10].

In conclusion, the thoracic type II endoleak is treatable with percutaneous CT-guided embolization, even in a case of massive hemoptysis.

**Ethical Approval:** All the study procedures were performed in accordance with the ethical standards of the approving institutional and/or national research committee and the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from the patient prior to the study.

**Conflict of interest:** The authors declare that they have no conflicts of interest to report.

**References**