


Failed transcatheter pulmonary artery embolization in a patient suffering from massive hemoptysis after thoracic endovascular aortic repair

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ABSTRACT: An emergency thoracic endovascular aortic repair (TEVAR) with zone 2 landing without revascularization of the left subclavian artery was performed due to the impending rupture of a distal arch aneurysm in an old patient presenting hemoptysis. Two months later, the patient had recurrent massive hemoptyses and continued after additional zone 0 TEVAR. The lung parenchyma was considered to be the bleeding source and transcatheter pulmonary artery embolization was performed, and the episodes of massive hemoptysis appeared to have ceased. However, the patient died of sudden recurrent massive hemoptysis 40 days later. Inflammation and/or infection of the lung parenchyma adjunct to the aortic aneurysm could be cause of fatal hemoptysis, and aggressive therapy such as lung resection should be considered in such patients.

KEYWORDS: Thoracic endovascular aortic repair, hemoptysis, pulmonary artery embolization

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Introduction

Hemoptysis in patients after thoracic endovascular aortic repair (TEVAR) is normally diagnosed with aortobronchial fistula or pulmonary parenchymal fistulation (ABPF) and treated with additional TEVAR or open surgical aortic repair.¹ However, if the aortic aneurysm is well excluded by stent-graft without endoleak, it is physiologically unlikely that there would be bleeding from the aortic wall through ABPF, causing hemoptysis. According to a certain patient cohort, hemoptysis post TEVAR should not be understood as ABPF, but rather as bleeding from lung parenchyma adjunctive to the aortic aneurysm—due to tissue inflammation, necrosis caused by mechanical compression, or local infection. Here, we present a case of transcatheter pulmonary artery embolization in a patient after TEVAR. Initially, the treatment was successful; however, the patient later died of recurrent massive hemoptyses. In this case report, the mechanisms of hemoptysis after TEVAR will also be discussed.

Case Report

Here, we present a 75-year-old man with a history of hypertension, paroxysmal atrial fibrillation, diabetes mellitus type II. One year prior, he suffered from a cerebral infarction and was treated in a rehabilitation hospital, whereafter he had regular outpatient appointments. For 1 month, he complained of general fatigue and weight loss. His laboratory data showed elevated inflammatory markers with a white blood cell (WBC) count of 11,000/ μ L and C-reactive protein (CRP) of 14.8 mg/dL. Moreover, he had anemia with hemoglobin of 10.5 g/dL, a decrease from 13.1 g/dL a month before. To find

the source of bleeding, gastroscopy and colonoscopy were done, but no gastrointestinal bleeding was found. Thereafter, chest plain computer tomography (CT) revealed a saccular aortic aneurysm of the distal arch with a diameter of 48 mm. For further examination, the patient was hospitalized immediately and a chest CT on the following day revealed enlargement of the aortic aneurysm to 54 mm in diameter (Figure 1A). Two hours after the CT examination, the patient suffered from massive hemoptysis. The patient was diagnosed with impending rupture of the aortic aneurysm and immediately transferred to our hospital. An emergency TEVAR was performed using (c-TAG 28 mm x 10 cm for distal and c-TAG 37 mm x 15 cm for proximal, Gore medical, Arizona, USA) with the right femoral approach, hereby the left subclavian artery had to be sacrificed (Figure 1B). Postoperative course was uneventful. Sulbactam/ampicillin sodium was only administered from pre-operation until the second postoperative day (POD) because the case was not initially treated as an infective aneurysm. The inflammatory markers rapidly improved with a WBC count of 5,770 μ L and CRP of 5.0 mg/dL, and the patient was transferred to the rehabilitation hospital where he was previously hospitalized.

In the rehabilitation hospital, the patient suffered from persistent slight fever and his blood culture was positive for *Streptococcus pneumoniae*. He was treated with a long-term oral administration of cefcapene pivoxil hydrochloride hydrate. Two months later, the patient suffered another sudden massive hemoptysis and was retransferred to our hospital. The CT examination revealed migration of the stent-graft and enlargement of the



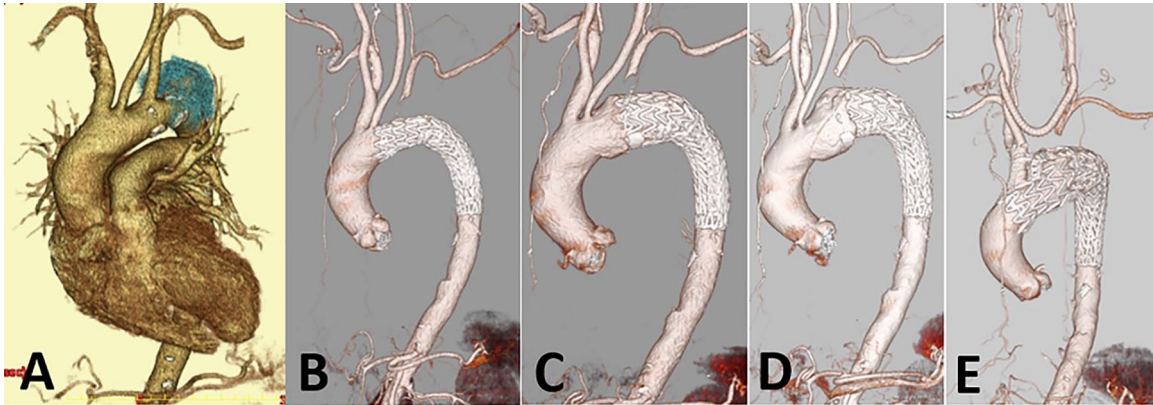


Figure 1. (A) Rapidly enlarging saccular aneurysm in the distal aortic arch. (B) Successful thoracic endovascular aortic repair without reconstruction of the left subclavian artery. (C) Stent-graft migration and enlargement of the aneurysm. (D) Rapid enlargement and further migration of the stent-graft. (E) Additional thoracic endovascular aortic repair TEVAR with 2 vessel debranching and chimney stenting in the brachiocephalic trunk.

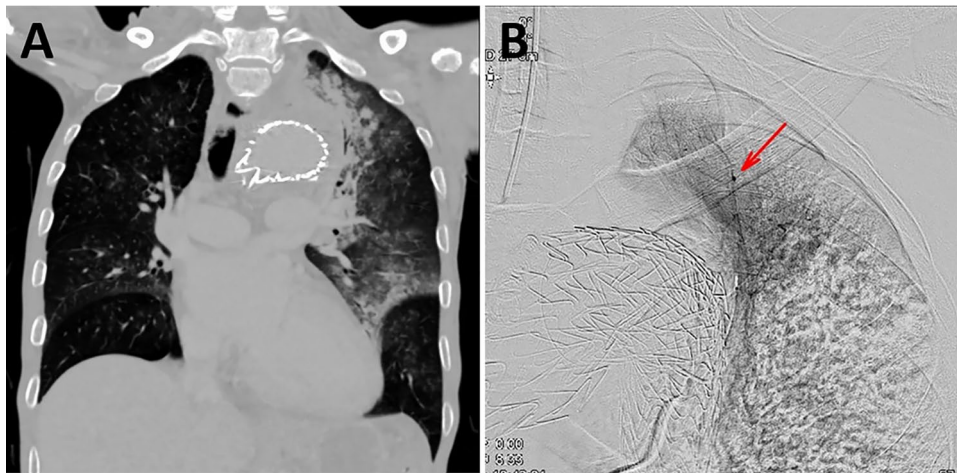


Figure 2. CT showed obvious lung consolidation in the left upper lobe adjunctive to the distal aortic arch (A). Angiography of the left pulmonary artery showed abnormal pooling of the contrast agent in the pulmonary artery branch of the left upper lobe (B, arrow).

aneurysm, without any sign of rupture (Figure 1C). His WBC count was 10,000/ μL and CRP was 9.3 mg/dL. Fluorodeoxyglucose positron emission tomography (FDG-PET) showed physiological accumulation typical to post TEVAR with a standardized uptake value of 4.8; infective aneurysm was negative according to the radiological report. Because the patient suffered hemoptysis once every 3 days, and the aneurysm rapidly enlarged in diameter within a matter of 7 days (Figure 1D), an additional TEVAR with debranching from the right common carotid artery to the left common carotid artery (Propaten 7 mm Gore medical, Arizona, USA) and proximal zone 0 landing (c-TAG 45 mm x 15 cm, Gore medical, Arizona, USA) and chimney stenting in the brachiocephalic trunk (E-Luminexx 12 mm x 80 mm, Bard, Arizona, USA) was performed (Figure 1E). However, the patient suffered from another massive hemoptysis on the first POD. The CT examination showed no endoleak, but an obvious lung consolidation in the left upper lobe adjunctive to the distal aortic arch. Because the aorta was considered to be a very unlikely source of bleeding, an

angiography of the left pulmonary artery was performed, revealing a pooling of the contrast agent in a branch of the pulmonary artery in the upper lobe (Figure 2). This branch was occluded using microcoils (VoltX, Boston Scientific, Marlborough, USA), thereafter, the bleeding ceased. Postoperative course was uneventful without hemoptysis nor fever. A gallium scintigraphy showed no accumulation around the aortic arch, and infective aneurysm was not suggested in the radiological report. Therefore, antibiotics were only administered in the perioperative period. The patient's WBC count normalized rapidly to 5990/ μL CRP decreased slowly to 7.6 mg/dL on the 14th POD. The patient was, again, transferred to the rehabilitation hospital on the 20th POD in stable general condition. However, he died of massive hemoptysis on the 40th POD in the rehabilitation hospital. Unfortunately, autopsy was declined.

Discussion

In this case, it is considered that the massive hemoptyses were not due to ABPF, but bleeding from lung parenchyma

adjunctive to the aortic aneurysm, because (1) there was no endoleak, (2) additional TEVAR without endoleak had no effect on reduction of the hemoptysis, and (3) hemoptysis ceased after transcatheter pulmonary embolization. However, the patient died of recurrent hemoptysis on the 40th POD. This case suggests that endovascular treatment is not enough for recurrent hemoptysis post TEVAR, and a more aggressive approach, such as lung resection may be necessary for this complex pathology.

ABPF is a rare but well-known complication after TEVAR and often results in fatal outcomes. In 2015, Czerny et al.¹ reported on 26 patients with ABPF from the European registry of endovascular aortic repair complications between 2001 and 2012 with a total caseload of 4680 TEVAR procedures in 14 centers. In their report, ABPF was defined as any communication between the thoracic aorta and the central airway or the pulmonary parenchyma post-TEVAR—but the communication was not directly proved in any of the patients, and endoleak was seen in only nine patients (35%). This study was a multi-center observational study, and it may be difficult to investigate each patient in detail, but it is possible that some of those 26 patients had no ABPF, but bleeding from lung parenchyma adjunctive to the aortic aneurysm, as the present case.

Considering the rapid expansion of the aneurysm in the initial phase of treatment, stent-graft migration with recurrent aneurysmal formation within only 2 months, and sustained systemic inflammation, it is retrospectively highly suspected that it may be a matter of mycotic aneurysm in the present case, although the patient had no fever and inflammation markers decreased rapidly after intervention. However, accurate diagnosis of mycotic aneurysm is sometimes difficult, even with advanced diagnostic modality such as FDG-PET, especially in patients with vascular/endovascular prosthesis.^{2,3} On the other hand, endovascular treatment for mycotic aortic aneurysm combined with long-term antimicrobial therapy has been recognized as a reasonable alternative in very high risk patients.⁴ Moreover, Maze et al.⁵ described that infected aortic grafts could be managed with long-term antimicrobial therapy and graft retention in selected patients. The present patient could have had another prognosis if he had received long-term antimicrobial therapy beginning in the first event. Therefore, we would like to consider

comprehensive and long-term antimicrobial therapy in such patients with suspected mycotic aneurysm.

To the best of our knowledge, this is the first report on pulmonary artery embolization in a patient suffering from hemoptysis post TEVAR, although our treatment plan ultimately failed. In the present case, hemoptysis was temporarily controlled by coil embolization of a pulmonary artery branch in the upper lobe, but the patient died of a recurred hemoptysis 40 days thereafter. This case suggests that endovascular treatment of the lung is not sufficient for treating the local lung inflammation adjunctive to the mycotic/infected aneurysm causing massive hemoptysis; lung resection with or without aortic repair may be the only way to treat such patients.

In conclusion, massive hemoptysis was not due to ABPF, but bleeding from lung parenchyma adjunctive to the aortic aneurysm in certain patient cohorts, especially cases where mycotic aneurysm is highly suspicious. In such patients, lung resection with or without aortic repair should be considered as a choice of therapy.

Author Contributions

Natsuya Ishikawa, Shinsuke Kikuchi and Kouhei Ishidou wrote this manuscript. Aina Hirofuji proofed this manuscript as a native English speaker. Sentaro Nakanishi, Hayato Ise and Naohiro Wakabayashi performed operations. Hiroyuki Kamiya supervised this case report.

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