Case Report

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Aortopulmonary fistula caused by an infected thoracic aortic false aneurysm rupturing after endovascular stent placement

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ABSTRACT: We report a case of 74-year-old man presenting with a rupture of a thoracic aortic false aneurysm after undergoing conventional total arch replacement for aortic arch aneurysm (62 mm) and endovascular stent placement for descending aortic aneurysm (70 mm). His chief complaints at the present admission were fever and sensation of dyspnea and we put him on a course of antibiotics for stent graft infection. However he died of massive hemoptysis. From a standpoint of autopsy findings, a thoracic aortic false aneurysm formed at the just proximal landing zone owing to type Ia endoleak, and simultaneously stent graft infection lead to make fistula formation between the false aneurysm and the lung. We examined ourselves that stent graft infection and aortopulmonary fistula caused by an infected thoracic aortic false aneurysm rupturing into the lung should be promptly treated such as complete removal of the stent and another revascularization in a reasonable period of time except if there are complications such as comorbid1ities or withholding of consent. We experienced and reported one rare case associated with a rupture of thoracic aortic false aneurysm caused by stent graft infection and the fistulization between the lung and the stent graft.

Keywords: Rupture, thoracic aortic false aneurysm, endovascular stent placement, graft infection, type Ia endoleak

1. Introduction

The initial experiences with endovascular treatment of thoracic aortic pathologies are promising, showing acceptable mortality and paraplegia rates (*1*). The indications for thoracic aortic endovascular repair include thoracic aortic aneurysms (TAA), acute and chronic expanding type B dissection, traumatic aortic rupture, and penetrating aortic ulcer (*2*). Although the number of thoracic endovascular aortic repair (TEVAR) is increasing rapidly, less is known about complications after TEVAR in contrast to abdominal endovascular aortic repair (AEVAR). Depending on the different aortic pathologies, procedure-related complications frequently occur. Serious complications include primary or secondary type I endoleak, retrograde type A dissection, stent collapse, and rupture with subsequent death. Series involving stent grafting of TAAs have shown that endoleaks occur in 3-29% (*3-7*), and about 50% of these are life threatening type I endoleaks with unchanged pressurized aneurysm sack. Meanwhile, Ducasse *et al*. demonstrated that the frequency of aortoiliac stent graft, which had been located by endovascular technique, infection in their study, *i.e.*, 0.43%, seems low and would indicate that stent grafts have a lower infection rate than that of prosthetic grafts used for conventional open repair (range 0.5-3%) (*8,9*). Although coexistence of these complications after TEVAR is rare, such pathological condition is fatal and the prompt therapeutic strategy for each individual case should be cautiously determined.

2. Case presentation

The case was a 74-year-old man whose chief complaints at the admission were fever and sensation of dyspnea. He had had hypertension for longer than 10 years and given β-blocker at the department of internal medicine of the hospital. In 1995, he presented with chest and abdominal pain. Chest computed tomography (CT) revealed an aneurysm of aortic arch of 52 mm in diameter. At first, medical conservative treatment to control blood pressure was chosen due to patient's wishes but it expanded to a diameter of 62 mm 5 years later, in 2000. The department of cardiovascular surgery of the hospital determined that it was an indication for surgery and performed a total aortic arch replacement. Postoperatively, another hospital found an aneurysm of descending aorta of up to 70 mm in diameter and carried out an endovascular stent placement in 2003.

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Since then, he had been followed up at our hospital and entered twice to treat hypertensive heart failure in 2004. He frequently underwent insertion and placement of a central venous catheter during these admissions. As of obstructive sleep apnoea syndrome which he developed in this period, continuous positive airway pressure was introduced and his systolic blood pressure was kept at around 140 mmHg by four antihypertensive medications, angiotensin II receptor blocker, angiotensin converting enzyme inhibitor, calcium channel blocker, and β-blocker. In March 2005, cold-like symptoms appeared and inflammation reaction continued high in spite of internal use of levofloxacin. Based on the left-sided accumulation of pleural effusion and expansion of the aortic aneurysm around the stent graft shown by chest X-ray (Figure 1), we considered that the patient had stent graft infection and hospitalized him for further investigation and treatment in April 2005. Examinations at admission revealed intensified inflammatory response, undernutrition, decreased renal function, and anemia associated with chronic inflammation (Table 1). Venous blood culture was positive for *Staphylococcus aureus*. As for the post-hospitalization progress, vancomycin hydrochloride and meropenem hydrate were given against stent infection probably caused by a previous central venous catheter placement, which reduced the patient's temperature and mitigated inflammatory response. However, on the 21st hospital day, his hemoglobin level suddenly dropped accompanying hematemesis and another onset of fever. In response to this, thoracic plain CT was performed on the 22nd hospital day, and it revealed the enlargement of the aortic aneurysm around the stent graft compared to that on the 12th hospital day (Figure 2). On the 22nd hospital day, large amounts of black watery stool appeared. Gastroscopy identified blood only in the trachea (Figure 3), which indicated hemoptysis, not gastrointestinal bleeding. The patient then developed aspiration pneumonia and received antibiotic treatment. On the 34th hospital day, the second massive hemoptysis appeared and caused airway obstruction and cardiac arrest. After receiving cardiopulmonary resuscitation, he was put under mechanical ventilator and given proper blood transfusion. On the 36th hospital day, he died of the third massive hemoptysis. Reoperation was not performed on request of the family during the admission. As for the result of autopsy, the aorta showed saccular dilatation macroscopically (Figure 4). Thinning of wall and massive infiltration of neutrophils were identified especially in the area where the stent was placed. Therefore, it was considered that the massive hemoptysis which was the immediate cause of death was led by a ruptured false aneurysm with a fistula opened to the lung parenchyma. The false aneurysm was located between the graft which was placed at the total aortic arch replacement and the stent which was later placed at TEVAR, and was formed probably through type Ia endoleak at the just proximal end of the stent. It seemed more likely that the proximal edge of the stent graft made contacts to the native aorta and subsequently endoleak occurred. Inflammation and infection at the site then caused formation of the fistula. Fistulization was microscopically confirmed in the adhesive area between the false aneurysm and the lung (Figure 5).

3. Discussion

Endovascular stent grafts are evolutionary medical devices that blend the vessel wall fixation properties of

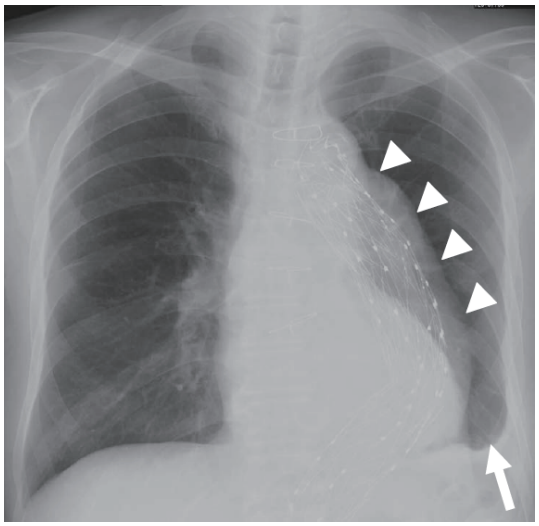

Figure 1. Chest X-ray on admission. Chest X-ray showed left-sided accumulation of pleural effusion (white arrow) and expansion of the aortic aneurysm around the stent graft (white arrowhead).

Table 1. Blood test findings on the present admission

Index	Measured value (Reference value)
Alb	2.2 g/dL (3.9-4.9 g/dL)
T-bil	0.3 mg/dL (0.2-1.0 mg/dL)
AST	27 IU/L (10-40 IU/L)
ALT	20 IU/L (5-45 IU/L)
BUN	36 mg/dL (7.2-20.0 mg/dL)
Cre	1.7 mg/dL (0.5-1.1 mg/dL)
Na	137 mmol/L (136-145 mmol/L)
K	3.6 mmol/L (3.6-4.8 mmol/L)
Cl	104 mmol/L (99-109 mmol/L)
LDH	122 IU/L (120-245 IU/L)
ALP	359 IU/L (104-338 IU/L)
γ-GTP	90 IU/L (16-73 IU/L)
CRP	16.4 mg/dL (< 0.3 mg/dL)
WBC	14,000 μL (3,100-9,500 μL)
Hb	9.4 g/dL (13.5-16.9 g/dL)
Plt	29.6×10^4 /μL (15.1-34.9/μL)
Fe	13 μg/dL (64-187 μg/dL)
Ferritin	404 mg/dL (27-320 ng/mL)
PT	76.9% (70-130%)
APTT	38.0 sec (24-38 sec)

Values in parentheses are normal ranges in our institution.

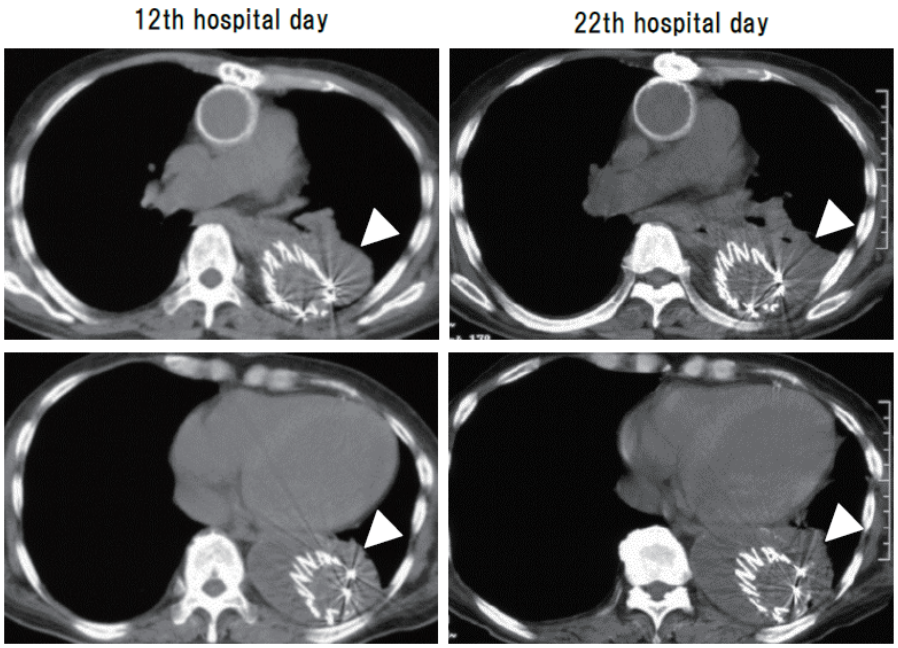

Figure 2. Changes of the thoracic CT findings during admission. The comparison between the findings of plain thoracic CT at 12th (left sided) and 22th hospital day (right sided) revealed that the aortic aneurysm around the stent graft had enlarged with time (white arrow head).

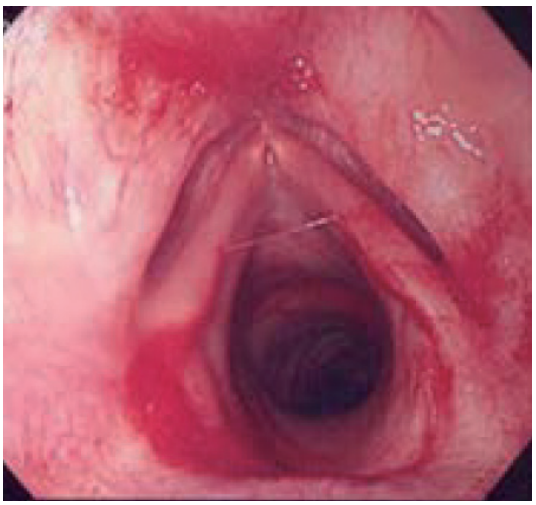

Figure 3. Gastroscopy findings at first hemoptysis. Gastroscopy revealed hemorrhage from the respiratory tract.

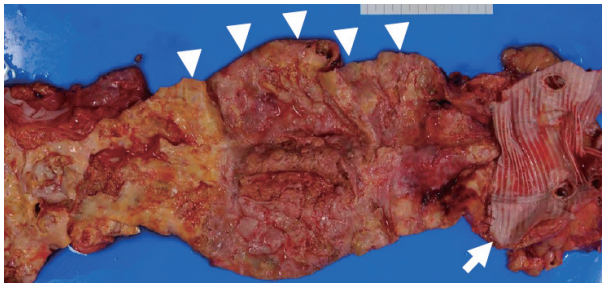

Figure 4. Macroscopic finding of descending aorta at autopsy. The autopsy specimen of the descending aorta, in which the stent graft placed at TEVAR had already been removed, showed saccular dilatation and thinning of the wall (white arrow head) at the distal side to the graft (white arrow), which had placed at the total aortic arch replacement. Right, cranial; left, caudal.

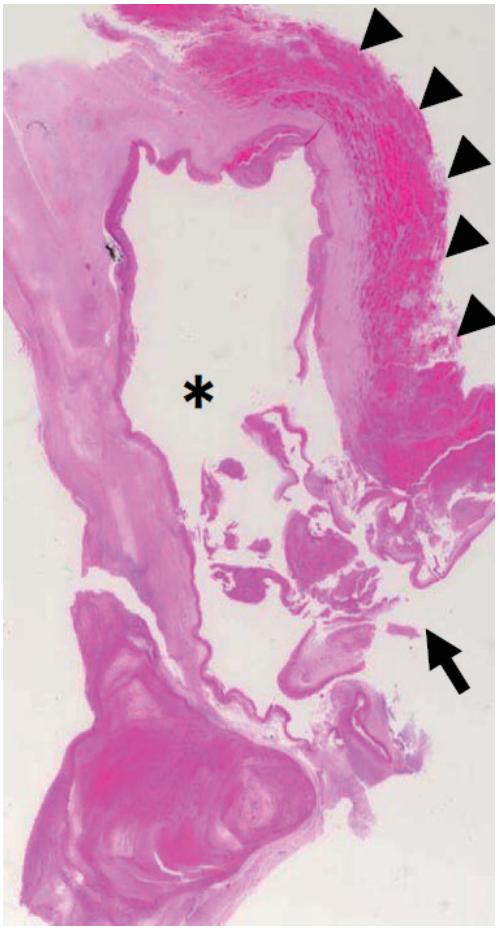

Figure 5. Microscopic finding of fistula detected between pulmonary parenchyma and aortic false aneurysm. Histological findings revealed the fistulization (black arrow) between the aortic false aneurysm (asterisk) and pulmonary parenchyma (black arrow head). H&E, ×100.

metallic intravascular stents with the arterial conduit properties of prosthetic vascular grafts. Endovascular stent placement has various advantages compared to surgical treatment, such as being noninvasive and low risks of spinal cord ischemia and bleeding. One of the first proposals for a minimally invasive intraluminal bypass was included with the initial clinical developments of catheter-based vascular intervention in the early 1960s (*10,11*). The landmark work by Dotter *et al*. using arterial angioplasty and vascular stents suggested the application of these newly developed devices to the treatment of traumatic arterial injuries and aneurysms (*12*). Marin *et al*. described that the overall technical success rate, which demonstrated free from major complications or major reinterventions, of TEVAR and AEVAR for TAA and abdominal aortic aneurysm was 85.1% in total of 817 patients over 10 years experience at one institution (*13*). However, immediately following TEVAR, 15% of patients had type I or III endoleaks and 10% had type II endoleaks (*13*). Additionally Alimi *et al*. emphasized the need for lifelong surveillance because the possibility for developing a late failure with type I endoleak of a previously excluded aneurysm was not negligible (*14*). Accurate detection and classification is essential for the proper management since the method of endoleak treatment is determined by the different source. High-pressure leaks (type I and type III) require urgent management because of the relatively high short-term risk of sac rupture. Once detected, endoleaks warranting correction are usually treated by endovascular route for poor surgical patients (*15*). A variety of techniques including extension endografts or cuff, balloon angioplasty, bare stents, and a combination of transvascular and direct sac puncture embolization techniques has allowed to treat the vast majority of these endoleaks without conversion to open surgical repair.

On the other hand, aortic stent graft infection is an uncommon complication and little is known about the general features and potential risk factors for aortic stent-graft infection, thus treatment is administered on a case-by-case basis with no consensus guidelines. Ducasse *et al*. demonstrated that more than half of patients (35/56, 62.5%) revealed stent graft infection at > 4 months after endovascular stent placement (*8*). They also showed that the offending microorganism was identified in 67.7% (44/65) and especially *Staphylococcus aureus* occupied the majority of patients (24/44, 54.5%) (*8*), in analogy with the present case. The treatments of stent graft infection could be grouped into conservative treatment, including using antimicrobial therapy and effusion drainage, and surgical treatment involving stent graft excision followed by aortic reconstruction. Ducasse *et al*. described that mortality of stent graft infection was 18% (11/61) overall, 36.4% (4/11) after conservative treatment, and 14% (7/50) after surgical treatment ($p = 0.086$). Despite a low infection rate, the associated mortality rates are extremely high and the morbidity rate, even with aggressive surgical interventions, is also high (*16*). Nevertheless, when stent infection occurred, complete removal of the stent graft and another revascularization should be advisable if feasible.

In the present case, it was certain that an aortopulmonary fistula occurred by an infected thoracic aortic false aneurysm rupturing into the lung. However, the trigger of enlargement of the thoracic aortic false aneurysm might be not only stent graft infection but also type Ia endoleak from the result of autopsy. The type Ia endoleak and the stent graft infection simultaneously might occur about 2 years after from TEVAR. Acctually etiologies of the aortic false aneurysms were reported to include leakage from surgical or endovascular placed graft, penetrating atherosclerotic ulcer, pneumonia, perforated esophageal ulcer, and mycotic aneurysm *(17)*.

Aortopulmonary fistula caused by an infected thoracic aortic false aneurysm rupturing after endovascular stent placement has rarely been reported and we should bear in mind that it is possible and unusually-serious. Although his family did not prefer the invasive treatment such as reoperation and reintervention, effective treatment should be promptly administered on a case-by-case basis. Further accumulation and elucidation of clinical cases including these complications are significantly required so as to overcome them in the future.

References

1. Leurs LJ, Bell R, Degrieck Y, Hobo R, Lundbom J; EUROSTAR; UK Thoracic Endograft Registry collaborators. Endovascular treatment of thoracic aortic diseases: Combined experience from the EUROSTAR and United Kingdom Thoracic Endograft registries. J Vasc Surg. 2004; 40:670-679.
2. Leurs LJ, Harris PL, Buth J. Secondary interventions after elective endovascular repair of degenerative thoracic aortic aneurysms: Results of the European collaborators registry (EUROSTAR). J Vasc Interv Radiol. 2007; 18:491-495.
3. Grabenwoger M, Fleck T, Ehrlich M, Czerny M, Hutschala D, Schoder M, Lammer J, Wolner E. Secondary surgical interventions after endovascular stent-grafting of the thoracic aorta. Eur J Cardiothorac Surg. 2004; 26:608-613.
4. Parmer SS, Carpenter JP, Stavropoulos SW, Fairman RM, Pochettino A, Woo EY, Moser GW, Bavaria JE. Endoleaks after endovascular repair of thoracic aortic aneurysms. J Vasc Surg. 2006; 44:447-452.
5. Ueda T, Fleischmann D, Dake MD, Rubin GD, Sze DY. Incomplete endograft apposition to the aortic arch: Bird-beak configuration increases risk of endoleak formation after thoracic endovascular aortic repair. Radiology. 2010; 255:645-652.
6. Grisafi JL, Boiteau G, Detschelt E, Potts J, Kiproff P, Muluk SC. Endoluminal treatment of type IA endoleak with Onyx. J Vasc Surg. 2010; 52:1346-1349.
7. Shah AA, Barfield ME, Andersen ND, Williams JB, Shah JA, Hanna JM, McCann RL, Hughes GC. Results of thoracic endovascular aortic repair 6 years after United

States Food and Drug Administration Approval. Ann Thorac Surg. 2012; 94:1394-1399.
8. Ducasse E, Calisti A, Speziale F, Rizzo L, Misuraca M, Fiorani P. Aortoiliac stent graft infection: Current problems and management. Ann Vasc Surg. 2004; 18:521-526.
9. Jackson MR, Clagett GP. Aortic graft infection. In: Decision Making in Vascular Surgery (Cronenwett JL, Rutherford RB, eds.). WB Saunders, Philadelphia, USA, 2001; pp. 186-191.
10. Fogarty T, Cranley J, Krause R, Strasser ES, Hafner CD. A method for extraction of arterial emboli and thrombi. Surg Gynecol Obstet. 1963; 116:241-244.
11. Dotter CT, Judkins MP. Transluminal treatment of arteriosclerotic obstruction. Description of a new technic and a preliminary report of its application. Circulation. 1964; 30:654-670.
12. Dotter CT. Transluminally-placed coil-spring endarterial tube grafts. Long-term patency in canine popliteal artery. Invest Radiol. 1969; 4:329-332.
13. Marin ML, Hollier LH, Ellozy SH, Spielvogel D, Mitty H, Griepp R, Lookstein RA, Carroccio A, Morrissey NJ, Teodorescu VJ, Jacobs TS, Minor ME, Sheahan CM, Chae K, Oak J, Cha A. Endovascular stent graft repair of abdominal and thoracic aortic aneurysms: A ten-year experience with 817 patients. Ann Surg. 2003; 238:586-595.
14. Alimi YS, Chakfe N, Rivoal E, Slimane KK, Valerio N, Riepe G, Kretz JG, Juhan C. Rupture of an abdominal aortic aneurysm after endovascular graft placement and aneurysm size reduction. J Vasc Surg. 1998; 28:178-183.
15. Cao P, De Rango P, Verzini F, Parlani G. Endoleak after endovascular aortic repair: Classification, diagnosis and management following endovascular thoracic and abdominal aortic repair. J Cardiovasc Surg (Torino). 2010; 51:53-69.
16. Numan F, Gulsen F, Solak S, Cantasdemir M. Management of endograft infections. J Cardiovasc Surg (Torino). 2011; 52:205-223.
17. Maruyama K, Ishiguchi T, Kato K, Naganawa S, Itoh S, Sakurai T, Ishigaki T. Stent-graft placement for pseudoaneurysm of the aorta. Radiat Med. 2000; 18:177-185.

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