Aneurysm formation at both ends of an endograft associated with maladaptive aortic changes after endovascular aortic repair in a healthy patient

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Abstract
We report a case in which saccular aneurysms formed at both ends of an endograft that exhibited maladaptive aortic changes after endovascular aortic repair (TEVAR) for a distal aortic arch aneurysm. A follow-up computed tomography (CT) scan performed at 6 months after the TEVAR detected a small saccular aneurysm at the distal edge of the endograft. At 10 months after the TEVAR, a new large aneurysm appeared at the proximal edge of the endograft. To prevent the latter aneurysm rupturing, total arch replacement with endograft fixation was performed. A CT scan obtained at 18 months after the TEVAR demonstrated that the aneurysm at the distal edge of the endograft had progressed and so we considered reintervention. Unfortunately, the patient died of intracranial haemorrhaging before the second procedure could be carried out. A histopathological examination of the aneurysm wall did not detect any significant background factors, such as connective tissue disease, inflammation or infection. The present case involved unexpected late complications, which might have been caused by changes in the form of the aorta after TEVAR.

Keywords: Thoracic endovascular aortic repair • Thoracic aorta

INTRODUCTION
Thoracic endovascular aortic repair (TEVAR) has become the standard treatment for descending thoracic aortic aneurysms and displays excellent postoperative mortality and morbidity rates [1]. Although the mid-term results of TEVAR seem to be satisfactory, late complications such as endoleaks, stent graft collapse and retrograde type A dissection have been reported [2, 3].

Herein, we report a case in which aneurysms formed at both ends of an endograft after endovascular aortic repair in a patient without vascular anomalies.

CASE REPORT
A 66-year old male was admitted to our institution for a thoracic aortic aneurysm in the distal aortic arch (Fig. 1A arrow). The patient was afebrile, but did not display elevated inflammatory marker levels during a preoperative blood examination (leucocyte count: 6500/μl, C-reactive protein: 0.02 mg/dl).

Since the proximal landing zone was sufficient (length: 20 mm), endovascular repair (TEVAR) was selected for this case. A Gore-TAG thoracic endoprosthesis (TGT3115, W.L. Gore and Associates, Flagstaff, AZ, USA) was inserted along an extra-stiff guidewire (COOK, Bloomington, IN, USA) and deployed into the distal arch. Postoperative computed tomography (CT) angiography performed 5 days after the TEVAR demonstrated that the endograft displayed good conformity and there were no endoleaks (Fig. 1B).

Three months after the TEVAR, a follow-up CT revealed the development of a small aneurysm along the greater curvature of the aortic arch at the distal edge of the endograft, associated with aortic reforms (Fig. 1C arrow) so we continued to carefully observe the patient. At 10 months after the TEVAR, a new large saccular aneurysm unexpectedly appeared at the proximal edge of the endograft (Fig. 2A and B). During the post-TEVAR follow-up period, the patient did not develop fever, and his serum inflammatory marker levels remained low (white blood cell count: 5100/μl, C-reactive protein: 0.66 mg/dl).

As the latter aneurysm seemed to be at high risk of rupture, total arch replacement was considered urgently. Through median sternotomy, a cardiopulmonary bypass was started via right axillary and right femoral perfusion with bicaval venous drainage. We opened the aneurysm under hypothermic circulatory arrest. A 28-mm Dacron graft (Triplex, Terumo, Tokyo, Japan) was inserted into the proximal end of the endograft as an elephant trunk and attached to the native aorta and the endograft using running sutures. Aortic arch reconstruction was performed using a four-branched Dacron graft (Triplex), and a distal anastomosis was made in the elephant trunk graft. After the graft replacement, we weaned the patient off the cardiopulmonary bypass without complications. He was discharged from hospital 21 days after the operation without any complications. A histopathological examination of the wall of the excised aneurysm revealed a thickened intima...
together with hyalinosis (Fig. 2D black arrow) and calcification, thinning of the collagen fibres in the media (Fig. 2D white arrow) and fibrosis of the adventitia accompanied by lymphocyte and plasma cell invasion. Also, the regions of the aortic wall outside of the aneurysm did not exhibit medial cystic necrosis or collagen disruption (Fig. 2E), which was not compatible with connective tissue disease, inflammation or infection.

A CT scan obtained at 18 months after the TEVAR revealed that the aneurysm at the distal edge of the endograft was slowly progressing (Fig. 2F). Reintervention was planned; however, the patient died of intracranial haemorrhaging (subcortical haemorrhage) 10 months after the total arch replacement. Intracranial CT angiography was performed, but no intracranial aneurysms or vascular malformation were detected.

**DISCUSSION**

The bird beak configuration (which is defined as poor conformity between the proximal end of an endograft and the aortic arch resulting in a wedge-shaped gap between the device and the aortic wall) is reported to be a significant risk factor for type Ia endoleaks [4]. Therefore, in the TEVAR procedure performed in the present case, we selected a straight region as the landing zone to avoid intimal injuries and the formation of the bird beak configuration at the edge of the endograft.

The present case was regarded to display a favourable aortic curvature for TEVAR. An early postoperative CT scan taken just after the implantation of the endograft demonstrated that it conformed well to the curvature of the aorta.

At first, we suspected that the aneurysms formed because the endograft was too big; however, after re-examining the patient with CT, we confirmed that the chosen graft was an appropriate size according to the manufacturer’s instructions (31 mm endograft inserted into 28 mm native aorta).

Also, we retrospectively compared the change in the curvature of the aortic arch on sagittal axis CT. As a result, we found that the curvature of the aortic arch had straightened within 3 months of the implantation of the endograft (Fig. 1A–C). The distal aneurysm formation began to progress after this change.
From these findings, we hypothesized that the forces responsible for the straightening of the curvature of the aortic arch might have caused aneurysms to form at both ends of the endograft. A previous report demonstrated that the TAG endograft is associated with a lower incidence of retrograde type A dissection [2] compared with the TX2 and Talent endografts, which suggests that lower levels of radial force are generated at the edge of the TAG. However, in the present case flaring of the edge of the endograft might have contributed to the patient’s intimal injury. A clinical examination of the aorta performed after the TEVAR procedure suggested that the aortic wall was fragile, which is commonly observed in patients with connective tissue disease. However, the patient had no relevant family history or major symptoms of connective tissue disease. In addition, a histopathological examination of the aneurysms did not demonstrate any anomalies in the media of the aortic wall. In patients with connective tissue disease, the long-term outcomes of TEVAR are not satisfactory because of endoleaks and the progression of aortic disease [5]. In such cases we should carefully examine the pathology of the aneurysm before considering the treatment options. The present case suggests that certain unknown aortic pathologies might be contraindicated for endovascular repair.

In conclusion, lifelong follow-up is mandatory after TEVAR, and surveillance based on sagittal view CT, which allows clinicians to see the alignment of the endograft, might be useful for predicting unexpected aortic changes. The development of thoracic endoprostheses with increased conformability would also help to avoid the complications we experienced.

Conflict of interest: none declared.

REFERENCES