The bronchial obstruction as a complication of endovascular repair of aortic pseudoaneurysm in Behçet’s disease

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Abstract: Behçet’s disease (BD) is an autoimmune disorder affecting multiple organs. Aortic pseudoaneurysm is the most catastrophic lesion in BD. This lesion type is considered as a complicated and challenging pathology by surgeons because of the technical operative difficulties and frequent recurrence. So, the endovascular repair of inflammatory aortic pseudoaneurysm has been used as an alternative to open surgical repair. It is particularly important in patients who are high-risk surgical candidates because of comorbidities. In this report, we present a case and treatment of bronchial obstruction, which caused progressive dyspnea after endovascular repair of aortic rupture, in patient with known history of BD.

Keywords: endovascular repair, bronchial obstruction, Behçet’s disease, pseudoaneurysm, autoimmune disorder

Case Report

A 46-year-old man with hemoptysis and palpitation presented to our emergency department. He had a history of Behçet’s disease (BD) for a period of 10 years, and 3 weeks before his admission, he was operated for lumbar hernia. Before the operation, he had discontinued immunosuppressive medications, which included steroids and azathioprine. A contrast-enhanced thoracic computed tomography (CT) was performed for a possible pulmonary artery aneurysm rupture and other emergent chest disease. CT scan showed a 8.0 × 9.5 cm aortic pseudoaneurysm with contained rupture in the aortic arcus, and the diameter of proximal landing zone was 29 mm, but aortobronchial fistula was not clearly seen (Fig. 1A, B). The patient was emergently taken to the interventional unit, and the pseudoaneurysm was excluded with implantation of the endovascular Valiant stent graft (30 × 100 mm; Medtronic, Santa Rosa, CA, USA) via the left femoral approach under conscious anesthesia, and there was no endoleak on the angiography (Fig. 2A). After 6 h, he complained of dyspnea. A chest roentgenogram taken showed that the left lung area was completely opacified (Fig. 3). Chest CT showed that the pseudoaneurysm growth in endovascularly repaired aorta was progressive and obstructed the left main bronchus (Fig. 4A). After routine preparation, a fully covered self-expanding stent (18 × 14 × 14 carinal Y stent – Novatech Dumon Y stent; Boston Medical Products, Westborough, MA, USA) was successfully placed in trachea and main bronchi (Fig. 4B). After this procedure, a second Valiant stent graft (34 × 100 mm) was implanted more proximally into the first endovascular graft with no endoleak (Fig. 2B). Finally, the patient was discharged to home in a stable condition with antithrombotic and immunosuppressive drugs. A CT angiography performed 3 months later showed regression of hematoma and the patient had no complaints.

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Discussion

BD is an autoimmune disorder affecting multiple organs. Aortic involvement occurs in one-third of BD patients with vascular disease [1], and it is more common in men. The first presentation of the disease occurs between the age of 20 and 40 years. Vascular inflammation is considered to be the main pathophysiologic lesion underlying vascular disease in BD. Aortic pseudoaneurysm is the most catastrophic lesion in BD. The incidence of rupture and death is not certain, but these lesions are generally considered to be of high risk. Surgical treatment is regarded essential; however, aortic pseudoaneurysm in BD is considered as a complicated and challenging pathology by surgeons because of the technical operative difficulties and frequent recurrence. Several studies describe the complications of open surgical treatment.

Fig. 1. (A) CT shows aortic pseudoaneurysm with contained rupture (asterisk). (B) 3D CT reconstructed view of the pseudoaneurysmal enlargement of the aorta (star)

Fig. 2. (A) Angiography shows the first performed thoracic endovascular aortic repair (TEVAR) stent graft. (B) Angiographic image of the second endovascular procedure, which was performed more proximally than the first procedure

Fig. 3. Chest roentgenogram reveals the opacified left hemithorax
for aortic pseudoaneurysm [2–4]. Aneurysm-related mortality with surgical procedures is 10–30% and recurrent pseudoaneurysm at an anastomotic site develops in 10–50% of patients.

Endovascular repair of inflammatory aortic pseudoaneurysm has been used as an alternative to open surgical repair since the first successful endovascular intervention performed by Dake et al. in 1994 [5], and it has the advantages of reduced invasiveness, fewer complications, and faster recovery than after surgical repair. These benefits are particularly important in patients who are high-risk surgical candidates because of comorbidities. At long-term follow-up, endovascular treatment of aortic pseudoaneurysm is safe and effective [6–11], and it has been found to have higher survival rates [7–9]. We performed thoracic endovascular graft implantation on emergency condition, and we used the optimal graft stent that could be easily found. Hence, that might be undersized, and it was partly responsible for pseudoaneurysmal sac enlargement after the procedure. Second graft implantation was successfully performed, thanks to its placement and sizing. In BD, immunosuppressive treatment reduces the complications of vascular involvement (rupture, thrombosis, etc.) and may result in regression of lesions [7]. So, in this case, we believe that discontinuation of the immunosuppressive medication with added operative stress after the lumbar hernia operation could be related to aortic rupture.

Previous studies conclude that immunosuppressive therapy should be started in patients with arterial disease before endovascular treatment or surgery and should be continued after the procedure to avoid the early and late post-procedure complications [3, 8, 9].

In this case, bronchial obstruction with pseudoaneurysm was solved by placing an endobronchial stent. In the setting of airway obstruction, endobronchial stenting can be life-saving to permit respiratory recovery, as well as long-term bronchial patency. The main indication for bronchial stent deployment is unresectable malignancy, but it is also used for resolving extrinsic bronchial obstruction caused by aneurysm [10].

**Conclusion**

In this report, we presented a case with aortic pseudoaneurysm in BD and implicated the following three points: (1) the safety and effectiveness of endovascular repair of the aortic pseudoaneurysm in patients with BD, (2) the importance of immunosuppressive drug continuation before and after the repair (both endovascular and surgical), and (3) lifesaving role of endobronchial and endovascular hybrid procedures in the management of pseudoaneurysm-related complications.

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