Endovascular Treatment of Left Subclavian Artery Pseudoaneurysm after Clavicle Fracture in an Elderly Adult with a 40-Year History of Behçet’s Disease

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Behçet’s disease (BD) is a rare chronic systemic vasculitis of unknown etiology that presents in young adults. We report a very rare case of a left subclavian artery pseudoaneurysm identified after a left clavicle fracture in a 70-year-old man with a 40-year history of intestinal BD. The patient received fixation by banding to treat a fracture after falling from a bed about 5 months previously, and he was referred to our hospital due to the finding of a left subclavian pulsatile mass. As enhanced computed tomography showed a left subclavian artery pseudoaneurysm, it was repaired using an endovascular stent graft.

Keywords: endovascular treatment, Behçet’s disease, subclavian artery pseudoaneurysm

Introduction

Behçet’s disease (BD) is a rare chronic systemic vasculitis of unknown etiology characterized by recurrent oral aphthous ulcers, genital ulcers, uveitis, and skin lesions in young adults aged 20 to 40 years.1–3 Postoperative complications, such as graft occlusion or anatomic pseudoaneurysm formation, have been frequently reported after open surgical repair in BD patients.4,5 Moreover, traumatic subclavian arterial rupture is a rare complication of blunt chest trauma.6 We report the successful endovascular treatment of a left subclavian artery pseudoaneurysm identified after a left clavicle fracture in a 70-year-old man with a 40-year history of intestinal BD, which is a very rare case.

Case Report

A 70-year-old man with a 40-year history of intestinal BD was referred to our hospital due to a left subclavian pulsatile mass measuring 4 cm × 4 cm (Fig. 1A), incidentally found when the patient received fixation previously by banding to treat a left clavicle fracture after falling from a bed about 5 months ago (Fig. 1B). Although he had recurrent oral aphthous ulcers and genital ulcers in his 30s, he had no BD-specific symptoms. As he also had a medical history of rheumatoid arthritis as well as chronic hepatitis C and aortic regurgitation, he was administered prednisolone (5 mg/day), colchicine (1 mg/day), and methotrexate (4 mg/day). The patient’s height was 157 cm and body weight was 46 kg. Upon arrival, the blood pressure was 176/81 mmHg and pulse was 75/min. Physical examination was unremarkable except for the left subclavian pulsatile mass. Laboratory examination showed: blood platelets, 6.6 × 10^4/µL; PT-INR, 1.35; fibrin degradation product (FDP), 44.4 µg/dL; red blood cells, 271 × 10^4/µL; hemoglobin, 9.4 g/dL; and cholinesterase, 80 IU/L. According to the disseminated intravascular coagulation (DIC) score determined by the Japanese Association for Acute Medicine,7 the DIC score was seven points. Enhanced computed tomography (CT) revealed a left subclavian artery pseudoaneurysm (Fig. 2A). As we were concerned about postoperative complications, such as graft occlusion or anatomic pseudoaneurysm formation, we decided on endovascular treatment for the present case. After selective angiography of the left subclavian artery via a right common femoral artery (CFA) showed bleeding from the left subclavian artery (Fig. 2B), we measured the diameter of the artery using intravascular ultrasound (IVUS). After an 8-mm × 60-mm endovascular stent graft (Fluensis Plus, Bard) was deployed (Fig. 2C), we confirmed adequate stent expansion and no dissection using IVUS. Finally, we dissected the right inguinal region to repair the CFA where the 9 Fr. sheath was inserted using 5-0 prolene.
After the endovascular treatment, the left subclavian pulsatile mass disappeared, while postoperative CT revealed no mass at the left subclavian artery. Although the blood platelets did not increase and PT-INR was still slightly elevated at around 1.3, FDP was markedly decreased and the DIC score was 5 points on the 8th postoperative day (Fig. 3). As PT-INR was slightly elevated and he had liver dysfunction, we did not administer anti-platelet therapy. Moreover, he continued to be administered prednisolone (5 mg/day), colchicine (1 mg/day), and methotrexate (4 mg/day) to control inflammation, and we confirmed to assess blood markers such as white blood cells (WBC) and C-reactive protein (CRP) as well as the presence/absence of BD-specific symptoms. The patient was followed up every 6 months, and the postoperative course remained uneventful at one year after treatment.

**Discussion**

BD is prevalent in Mediterranean countries. Turkey has the highest prevalence at 80 to 370 cases per 100 000 of the population. The prevalence in Japan, Korea, China, Iran, and Saudi Arabia ranges from 13.5 to 20 cases per 100 000, while that in Western countries is lower.\(^{1,3}\) The main symptoms are recurrent oral aphthous ulcers, genital ulcers, uveitis, and skin lesions, while the other symptoms are arthritis and epididymitis, and BD also involves the central nervous system, gastrointestinal lesions, and vascular lesions.\(^{1–3}\) Vascular BD includes arterial and venous thrombosis and aneurysm formation.\(^{1,3}\) The frequency of vascular BD was reported to be 3%,\(^{3}\) 9% in Japan, and 38% in Turkey.\(^{1,8}\) In the present case, after the patient had undergone intestinal resection 40 years ago, he was diagnosed with intestinal BD. Moreover, he was diagnosed with the left subclavian artery pseudoaneurysm when it was found incidentally during fixation to treat a left clavicle fracture about 5 months ago. Although we think that the history of BD had influenced the subclavian artery, and also prednisolone had worsened the condition and was related to the fragile artery and, thus, the pseudoaneurysm, to our knowledge, there has been no report of a similar case.
Due to the postoperative complications, such as graft occlusion or anatomic pseudoaneurysm formation, endovascular treatment of an aortic pseudoaneurysm in a BD patient has been reported.\(^9\,10\) In the present case, we also decided to perform endovascular treatment due to the possibility of complications. Even though we performed this treatment, in order to prevent complications, firstly, we measured the diameter of the artery using IVUS to select an appropriate stent graft, and secondly, we did not perform balloon dilatation to prevent recurrent aneurysm at the edge of the inserted graft. Moreover, we dissected and repaired the CFA where the 9 Fr. sheath was inserted to prevent the aortic pseudoaneurysm rather than solely for percutaneous pressure hemostasis. However, as an endovascular stent graft is not covered by health insurance in Japan, we hope to change the situation regarding health insurance coverage.

Traumatic subclavian arterial rupture is a rare complication of blunt chest trauma.\(^6\) Subclavian artery injuries are caused by either elongation or laceration mechanisms. Elongation is characteristically associated with blunt force applied to the anterior shoulder or clavicle, while laceration to the subclavian artery is due to bony fragments produced by a fractured first rib or clavicle.\(^10\) In the present case, the arterial injury occurred due to laceration. Although it is possible to suffer a stent graft fracture due to a fractured clavicle, the postoperative course remained uneventful at one year after treatment. Moreover, there have been many reports of endovascular treatment for subclavian arterial injuries,\(^6\) even in BD patients.\(^9\) The present case does not allow us to conclude that endovascular treatment may be a less invasive approach for a subclavian artery pseudoaneurysm in a BD patient; however, it is worth noting that a history of BD is an important consideration in patients with clavicle fracture.

**Conclusion**

We report a very rare case of a left subclavian artery pseudoaneurysm after a left clavicle fracture in a 70-year-old man with a 40-year history of intestinal BD. It is worth noting that a history of BD is an important consideration in patients with clavicle fracture.

**Disclosure Statement**

All authors declare that they have no conflict of interest.

**Author Contributions**

Study conception: K.N

Data collection: K.N

Analysis: K.N

Investigation: K.N, T.H

Writing: K.N

Funding acquisition: K.N

Critical review and revision: all authors

Final approval of the article: all authors

Accountability for all aspects of the work: all authors

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