Case Report

Urgent Surgical Management of Deep Femoral Artery Aneurysm in a Patient with Pre-Vasculo-Behcet Status

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We report a case of deep femoral artery (DFA) aneurysm associated with pre-vasculo-Behcet status. A 34-year-old man with a history of recurring oral and genital ulcers was admitted complaining of worsening left thigh pain over the previous 30 days. Computed tomography showed a left DFA aneurysm (60 mm × 70 mm), concomitant aneurysms in the popliteal and carotid arteries, and deep vein thrombosis. Active pre-vasculo-Behcet status was diagnosed, and DFA ligation was performed urgently. Remission was achieved with postoperative prednisolone and colchicine without vascular complications. DFA aneurysm and vascular pathologies were successfully managed by ligation surgery and medical therapy.

Key words: Behcet disease, aneurysm, deep femoral artery

Background

Behcet disease (BD) is a chronic, relapsing inflammatory illness that involves multiple organ systems, and its underlying etiology remains unknown. The clinical presentations include mucocutaneous, genital, ocular, skin, articular, gastrointestinal, neurological, and vascular involvement.1–3) The incidence of vascular involvement ranges from 12.8% to 35.1%,4,5) and various vascular pathologies have been reported to occur in BD patients. Arterial involvements, such as aneurysm and occlusion, can occur in BD patients, can lead to fatal complications, and are considered to be of clinical importance.6–8) Arterial involvements are usually treated surgically. There has been no clear consensus on the optimal timing of treatment; however, it was reported that postponing surgical intervention until remission may decrease the incidence of later vascular complications.6) We report here a rare case of large deep femoral artery (DFA) aneurysm in a patient with BD who underwent urgent surgical intervention while in the active phase of disease. This case was successfully managed by surgery and postoperative immune-suppressive therapy.

Case Presentation

A 34-year-old man was admitted to an emergency department of a district hospital, with a complaint of worsening left thigh pain. The symptoms occurred suddenly without traumatic events 25 days previously. The pain was temporarily relieved with the use of an analgesic agent; however, severe pain recurred, causing gait disturbance. Computed tomography (CT) scanning after admission showed a large aneurysm (60 mm × 70 mm) in the left DFA, a smaller aneurysm in the left popliteal artery (15 mm × 17 mm), and deep vein thrombosis in the left popliteal vein (Fig. 1). The patient was referred to our hospital for further evaluation and treatment. He had a history of recurring oral and genital ulcers since the age of 6 years. He had no family history of autoimmune diseases or BD. Physical examination on admission revealed a swollen left thigh with a palpable mass with tenderness. The popliteal artery and dorsal pedis artery were palpable, and there were no findings of leg ischemia. Laboratory tests showed an accelerated erythrocyte sedimentation rate of 82 mm/h and increased C-reactive protein level of 7.3 mg/l. His clinical manifestations did not fulfill the diagnostic criteria of the International Study Group for Behcet’s Disease clinical assessment, due to a lack of characteristic skin and ocular lesions.9) However, characteristic vascular pathologies strongly suggested the possibility of BD, and the lesion was diagnosed as pre-vasculo-Behcet status. He had not been treated with immune-suppressive agents. Thus, conservative medical therapy was considered as an initial therapy for the un-ruptured aneurysm to
achieve remission. However, the large painful aneurysm suggested a possible risk of impending rupture. The patient underwent urgent radical DFA ligation surgery. Under general anesthesia, the patient was placed in the supine position. A longitudinal incision was made along the sartorius. First, the common femoral artery, superficial femoral artery (SFA), and proximal DFA were located. Next, the second perforator of DFA, the main vessel in the distal side of the aneurysm, was identified in the space between the sartorius and the adductor longus. The DFA aneurysm was located adjacent to SFA. After ligation of the proximal DFA, the aneurysm was opened. The aneurysmal wall was thin and fragile, and a punched-out defect of the arterial wall was identified. The rupture site was sutured, and two branch vessels connected to the aneurysm were ligated. Finally, the second perforator was ligated, and complete hemostasis was confirmed. The operation time was 190 min. Histological findings showed the destructed integrity of the arterial wall with cell infiltration, which was consistent with a pseudoaneurysm (Fig. 2).

The patient did not show symptoms of ischemia in the left lower extremity postoperatively. Head contrast CT scanning showed a left carotid artery aneurysm of 10 mm in diameter (Fig. 1). Genetic investigation showed that the patient was positive for HLA-A26, but not HLA-B51.

Immune-suppression therapy with prednisolone (50 mg per day) and coumarin was started postoperatively. He responded well to the medical therapy, and his C reactive protein level returned to normal. The postoperative course was uneventful. The patient was discharged from the hospital on postoperative day 43. Maintenance doses of prednisolone (5 mg/day) and colchicine (0.5 mg/day) were started during the outpatient follow-up. Over 18 months of follow-up, the patient showed no evidence of vascular complications. Follow-up CT examination demonstrated that there was no recurrent pseudoaneurysm formation at the previous ligation sites, and the peripheral aneurysms had not expanded. Although the patient received anticoagulation therapy with warfarin, ultrasound examination showed that small deep vein thrombosis still remained in the left popliteal vein. Oral and genital ulcers were in remission.

Discussion

Vascular involvement is relatively rare as an initial clinical manifestation of BD. Kural-Seyahi et al. reported that, of 428 BD patients, more than 94% of patients showed oral and genital ulceration at the initial visit. On the other hand, only 20.6% of patients showed vascular involvement at the initial visit. Nevertheless, vascular involvement represents a major cause of mortality of BD patients, as well as central nervous system involvement or cancer. Its impact on clinical outcomes is high, and thus, understanding the clinical course and treatment effects is quite important for daily clinical practice. Vascular involvement tends to occur in male patients, and venous diseases are more common than arterial diseases. The most common vascular involvement is superficial or deep vein thrombosis, and subsequent fatal pulmonary embolism, although rare, has been reported. The other vascular involvements include arterial aneurysm, arterial stenosis or occlusion, inferior or superior vena cava thrombus, pulmonary artery aneurysm, and Budd–Chiari syndrome. The incidence of arterial involvement ranges from 1.0% to 7.0%. Furthermore, peripheral artery involvements are less common, with a reported incidence ranging from 0.5% to 4.3%. As shown in the present case, patients with arterial involvements tend to have multiple lesions and concomitant deep vein thrombosis. Aneurysm is more common than occlusion. Regarding site of occurrence, there have been some geographic variations in cohorts studied (Table 1). The femoral artery and abdominal aorta have been reported to be sites of predilection for aneurysm from the reports of Turkey and Korea. On the other hand, Fei et al. reported that, of 56 arterial lesions from the Chinese registry data, the most common site was lower extremity and abdominal aorta (n = 15 each), and only one patient showed a femoral lesion. DFA may have been included as a “femoral artery” in the previous studies; however, the details

![Fig 1. Computed tomography scan images. A: Axial view of the deep femoral artery aneurysm adjacent to the superficial femoral artery. B: Axial view of the popliteal artery aneurysm and deep vein thrombosis of the popliteal vein. C: Three-dimensional imaging of the left and right carotid artery showing an aneurysmal dilation in the origin of the left internal carotid artery. SFA: superficial femoral artery; DVT: deep vein thrombosis.](image-url)
regarding the treatment of DFA aneurysms associated with BD has not been reported. We consider that the current report can provide a road map for this rare, but challenging clinical condition.

The treatment strategy for a peripheral artery aneurysm associated with BD is determined by the anatomical location and clinical presentation, including rupture or impending rupture and the active- or remission-stage of disease. The surgical outcomes of arterial involvements are highly influenced by vascular complications based on the severe inflammatory nature of this disease. Koksoy et al. reported relatively unfavorable surgical outcomes of peripheral arterial involvements. Of 15 femoral artery surgeries (14 graft interpositions and one ligation) for BD patients with vascular involvement, four graft infections, six graft occlusions, and five anastomotic pseudoaneurysms occurred, leading to four amputations and six late deaths. Of note, an anastomotic pseudoaneurysm is a life-threatening complication, and weakening of the arterial wall caused by fulminant inflammation may lead to

**Fig 2.** Histological findings (A, C: hematoxylin-eosin stain, B: elastica van Gieson stain). Examinations under low-power view (A, B) indicate that the preserved arterial wall integrity (blue arrow) is shown in a limited area (the right side of red arrow). Arterial wall integrity is destructed in the remaining area with massive cell infiltration, which is consistent with a pseudoaneurysm. High-power view (×20) shows nonspecific inflammation with neutrophilic infiltrate.
anastomotic fragility. Kalko et al. reported their treatment policy for vascular BD. They recommended avoiding surgery in the active phase immunosuppression of the disease as a key to preventing later pseudoaneurysm formation. Management for arterial involvements associated with BD requires perioperative and postoperative comprehensive medical therapy to control the inflammation. Although the present case showed a large aneurysm with impending rupture, the aneurysm was fortunately located in DFA. Therefore, a simple ligation technique was possible even in active phase immunosuppression. Postoperative immune-suppression started with 50 mg/day prednisolone, and the amount was gradually decreased. Colchicine was added as outpatient medical therapy. These medications successfully controlled the inflammation of the disease without vascular complications or progression of the concomitant peripheral aneurysms. To prevent vascular complications, Kalko et al. advocated using a synthetic material, such as PTFE graft, because the inflammation may spread to autologous venous grafts. Maeda et al. reported the successful management of celiac artery aneurysm with extra-anatomical aorta-common hepatic artery bypass using e-PTFE graft. However, Koksoy et al. reported that the choice of graft material did not affect the outcomes. Thus, there has not been a consensus regarding the most suitable graft material for arterial involvements associated with BD.

An alternative treatment for arterial involvements is an endovascular approach. Kim et al. reported that 20 arterial aneurysms were treated with an endovascular technique after induction of remission by preoperative immunosuppression therapy. In all patients, a stent graft was successfully placed with acceptable later vascular complications including two stent graft occlusions and two access site pseudo-aneurysms. Although its indication is limited by anatomical factors and data regarding late outcomes are lacking, endovascular treatment may be a safe and less invasive modality for arterial pathology associated with BD. In cases in which ligation cannot be performed because of peripheral ischemia, the endovascular approach may be an alternative treatment option for arterial involvement associated with BD.

**Conclusion**

Although the patient was in the active phase of pre-vasculo-Behcet status, we successfully performed urgent ligation surgery for a large DFA aneurysm before impending rupture. Remission was achieved with postoperative prednisolone and colchicine without vascular complications or progression of the concomitant peripheral aneurysms.

**Disclosure Statement**

The authors have no conflict of interest to disclose.

**References**