


# Endovascular Treatment of the Ruptured Pseudoaneurysm of Common Femoral Artery in Behcet's Disease: Report of a Case with Four Years of Follow-Up

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## Abstract

Behcet's disease (BD) is a vasculitic condition that affects arteries and veins of all sizes. Arterial pseudoaneurysm is a rare but life-threatening complication of BD. We reported a 41-year-old man with 20 years history of BD and 5 years history of bilateral blindness. The patient presented with sudden onset of pain and swelling in his left groin and was diagnosed with the ruptured pseudoaneurysm of the left common femoral artery (CFA), which extended into the origins of superficial and deep femoral arteries. The patient was in the active phase of BD; therefore, he was initially treated with corticosteroids and cyclophosphamide pulse therapies and subsequently underwent stent graft placement. Post stenting angiography and postprocedural ultrasonography confirmed the successful exclusion of pseudoaneurysm without any evident complication. Four years of follow-up was uneventful.

## Keywords

Behcet's disease, pseudoaneurysm, endovascular treatment, rupture, stent graft

## Introduction

Behcet's disease (BD) is a vasculitic condition that affects arteries and veins of all sizes.<sup>1</sup> It usually manifests between the third and fourth decades of life.<sup>1,2</sup> This disease is characterized by recurrent oral aphthous ulcers, genital ulcers, cutaneous lesions, arthritis, and vascular, gastrointestinal, ocular, and neurologic manifestations.<sup>3</sup>

In a retrospective analysis of 2319 patients with BD in Turkey, vascular involvement was found in 14.3% of the patients, it was more common in men, and the veins were more commonly affected than the arteries.<sup>4</sup> Venous involvement tends to be in the form of venous occlusion; however, arterial involvement can be in both forms of arterial occlusion and arterial wall aneurysm formation.<sup>5</sup>

Although arterial involvement is uncommon among these patients, it is a major cause of morbidity and mortality.<sup>1</sup> The most commonly affected arteries are the aorta, carotid, iliac, femoral, popliteal, and pulmonary arteries.<sup>5</sup> Management of arterial aneurysms often involves a combination of medical treatment and surgical/endovascular treatment. Surgical and endovascular procedures are challenging in these patients due to the high risk of procedural complications, including arterial rupture, anastomotic aneurysms, graft infection, and graft

occlusion or stenosis.<sup>6</sup> When surgical or endovascular treatment is planned, the ideal approach is to perform the procedure in the remission phase of the disease.<sup>7</sup> However, urgent intervention is required in cases with a ruptured aneurysm or evident organ ischemia.

Herein, we present the case of a 41-year-old man with a medical history of BD and bilateral blindness who presented with sudden onset of pain and swelling in his left groin. The patient was diagnosed with a large ruptured pseudoaneurysm of the left common femoral artery (CFA), which extended into the origins of superficial and deep femoral arteries. The patient was successfully treated with endovascular stent graft placement.

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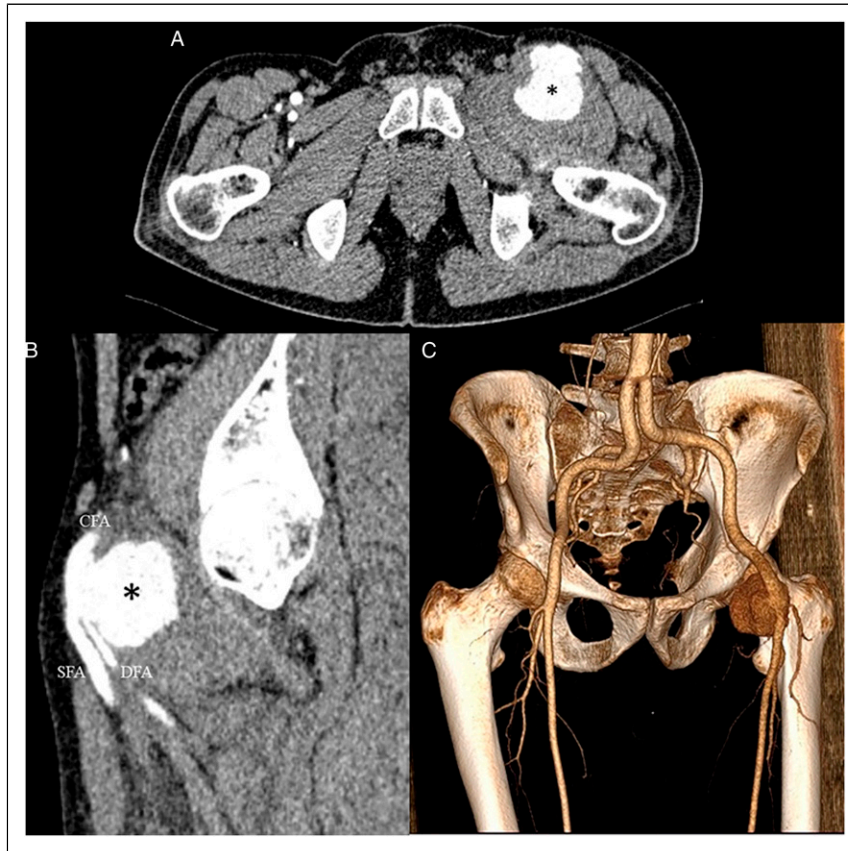
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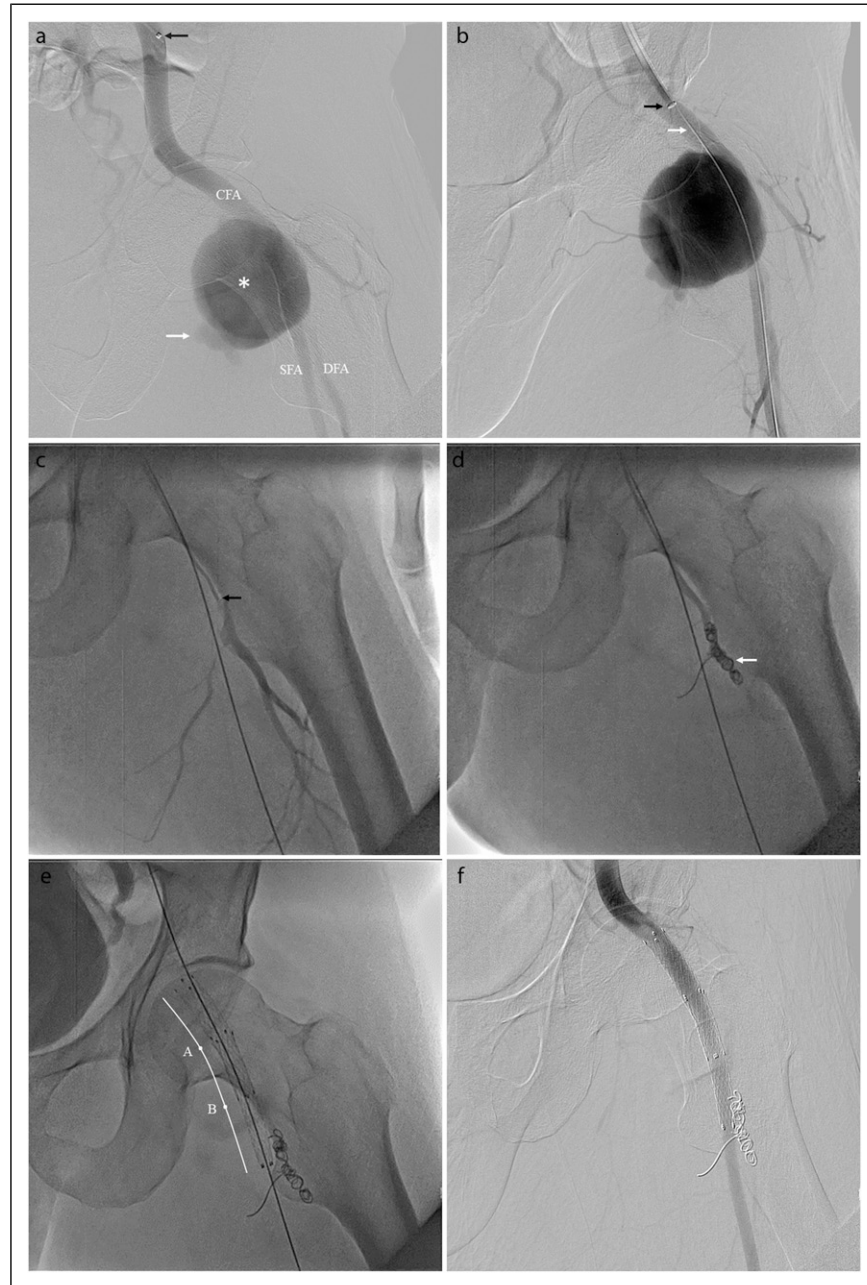
**Figure 1.** A 41-year-old man with Behcet's disease presented with sudden onset of pain and swelling in his left groin. (a) axial and (b) sagittal CT image demonstrates the saccular pseudoaneurysm and its surrounding hematoma (asterisk), common femoral artery (CFA), superficial femoral artery (SFA), and deep femoral artery (DFA). The pseudoaneurysm encompassed the origins of both SFA and DFA, and the neck of the pseudoaneurysm was measured 2.43 cm. (c) the 3-D volume rendering of pseudoaneurysm.

## Case Presentation

A 41-year-old Caucasian man with a medical history of BD presented to the emergency department with the complaint of pain and swelling in his left groin, which had suddenly started 1 day before. He denied any history of preceding trauma. The patient was diagnosed with BD and had developed bilateral blindness 20 and 5 years earlier, respectively. His past medical history was apparently unremarkable. He was initially prescribed prednisolone, cyclosporin, and aspirin; however, he stopped taking all the medications 5 years ago. His past surgical history was unremarkable, he did not smoke or use alcohol, and his family history was also unremarkable. Vital signs were within normal limits, and the physical examination was remarkable for a pulsatile mass in his left groin, which could be easily inspected and palpated. The right knee joint was also slightly swollen and warm. Bilateral popliteal, dorsalis pedis, and posterior tibialis pulses were symmetrically palpated (2+). Capillary refill, color, and temperature of the left lower limb were normal and identical to the right lower limb. Laboratory test results showed erythrocyte sedimentation rate (ESR) 72 mm/h (reference range up to 15 mm/h),

C-reactive protein (CRP) 1+, white blood cell (WBC) count 8900/microL, hemoglobin (Hb) 11.8 g/dL, and platelet count 380 000/microL. Ultrasonographic examination revealed a wide-necked saccular pseudoaneurysm of 53 × 50 × 62 mm on the posterior aspect of the common femoral artery (CFA) encompassing origins of both superficial femoral artery (SFA) and deep femoral artery (DFA), which was covered with hematoma. Blood flow was normal in the femoral and popliteal arteries. The pelvic and lower extremity computed tomography angiography (CTA) also confirmed the findings (Figure 1).

The patient was hospitalized and treated with prednisolone, colchicine, amlodipine, metoprolol, atorvastatin, and pantoprazole. The patient also received corticosteroids pulse therapy for successive 3 days and cyclophosphamide pulse therapy. On the seventh day of admission, ESR and CRP reached the normal limits, and endovascular treatment with stent graft placement was planned for the patient. The open surgical repair could not be an option in this patient as this patient was identified as high-risk for general anesthesia and open surgery by the anesthesiology service due to cardiopulmonary insufficiencies.



**Figure 2.** Stent graft placement in a 41-year-old man with Behcet's disease diagnosed with ruptured pseudoaneurysm of the left common femoral artery (CFA). (a) angiography demonstrates the CFA, superficial femoral artery (SFA), and deep femoral artery (DFA). Note the catheter tip (black arrow), pseudoaneurysm (asterisk), and contrast extravasation (white arrow). (b) a long sheath was positioned to the proximal CFA (black arrow), and a guidewire (white arrow) was advanced into the SFA crossing the pseudoaneurysm. (c) an additional guidewire was advanced into the DFA, and angiography of the DFA was performed. Note the catheter tip (black arrow). (d) proximal DFA was coil embolized (white arrow). (e) two 4 × 70 mm stent grafts (Fluency plus, Bard) were deployed longitudinally from the proximal SFA to the CFA with 3 cm of overlap. The area in-between the A and B points mark the overlap of the two stent grafts. (f) Completion angiography shows successful exclusion of the pseudoaneurysm without any sign of contrast extravasation or endoleak.

## Procedure Description

Retrograde puncture of the right common femoral artery (CFA) was performed under local anesthesia. Using the cross-over technique, access to the left external iliac artery (EIA) and left CFA was obtained with a 6-F Judkins Right catheter (JR, Cordis, USA). Angiography of the left EIA and left CFA confirmed the presence of a large pseudoaneurysm on the left CFA (Figure 2A). The pseudoaneurysm extended into the origins of both superficial femoral artery (SFA) and deep femoral artery (DFA) and demonstrated contrast extravasation (Figure 2A). A 0.035-inch Amplatz stiff guidewire (Cordis, USA) was advanced into the SFA crossing the pseudoaneurysm (Figure 2B), and an additional 0.035-inch standard hydrophilic guidewire (AqWire, Covidien, USA) was advanced into the DFA. Using a 5-F Vertebral catheter (Cordis, USA), angiography of the DFA was performed (Figure 2C), and the proximal DFA was embolized with two 5 cm × 5 mm coils (MR eye pushable coils, Cook Medical, USA) (Figure 2D). Coil embolization of the proximal DFA was performed to avoid the potential type II endoleak of the planned pseudoaneurysm repair. Subsequently, over the Amplatz stiff guidewire, two 7 × 40 mm stent grafts (Fluency plus, Bard, USA) were deployed longitudinally from the proximal SFA to the CFA with 3 cm of overlap (Figure 2E). The proximal landing zone of the proximal stent graft was positioned 1 cm below the acetabulofemoral joint line to decrease the risk of future stent graft fracture. Completion angiography showed successful exclusion of the pseudoaneurysm without any sign of contrast extravasation or endoleak (Figure 2F).

The post-procedural course was uneventful, and the pain and swelling gradually subsided. Examination with ultrasonography confirmed pseudoaneurysm exclusion without any evident complications. The patient was discharged on prednisolone, colchicine, aspirin, clopidogrel, atorvastatin, metoprolol, amlodipine, pantoprazole, and an additional dose of cyclophosphamide pulse therapy was planned. The patient was evaluated every 3 to 6 months with ultrasonography. Four years of follow-up was uneventful.

## Discussion

We reported a case of BD complicated with ruptured CFA pseudoaneurysm, which was successfully treated with endovascular stent graft placement. The treatment of this pseudoaneurysm was challenging from several aspects: (a) It had extended into the origins of both SFA and DFA (b) It was of a wide neck (measured 2.43 cm). (c) It had already been ruptured. (d) The patient was in the active phase of the BD. (e) open surgical repair could not be an option because of the patient's condition.

Peripheral artery aneurysms (PAA) in BD should be managed using a combination of medical (high-dose

corticosteroids and cyclophosphamide) and surgical (open surgery (OS) or endovascular treatment (ET)) treatments. However, cases with small and asymptomatic aneurysms which carry a low risk of rupture may be managed by exclusive medical treatment.<sup>8</sup> The choice between OS and ET has to be made based on the size and location of the aneurysm and also the team's experience.<sup>8</sup>

In a retrospective study<sup>6</sup> that included 27 PAAs (affecting aorta, iliac artery, femoral artery, popliteal artery, carotid artery, subclavian artery, renal artery, and superior mesenteric artery), 10 aneurysms were treated with ET, and 17 aneurysms with OS (12 with greater saphenous vein (GSV) grafts, and 5 with synthetic grafts). Although the difference between the complication rates was not statistically significant between the 3 methods, anastomotic aneurysms were only seen in GSV and synthetic grafts, and arterial rupture was only seen in 1 endograft. In addition, graft occlusion/stenosis was seen in 1 GSV, two synthetic grafts, and two endografts. Of note was that endograft stenosis or occlusions were more frequently seen when they were implanted in arteries near the joints (femoral and popliteal artery). It is speculated that, stent grafts positioned in close proximity of joints are predisposed to fracture due to the movements of the joint and stent graft's chronic outward forces.

In another retrospective study<sup>9</sup> that included 22 PAAs (affecting external iliac artery, common femoral artery, superficial femoral artery, and popliteal artery), no serious complication was recorded following the OS; however, 1 of their patients was complicated with stent graft fracture and thrombosis, four patients with stent graft occlusion, 1 patient with stent graft thrombosis, and two patients with stent graft stenosis. These complications were managed endovascularly except for the CFA stent graft fracture, which warranted conversion to open surgery. Based on these results, the authors recommended that CFA aneurysms should be treated with OS. However, there is another report in the literature on CFA aneurysm in BD which is treated with expanded polytetrafluoroethylene interposition graft. This patient was lately complicated with pseudoaneurysm recurrence at the site of surgery.<sup>10</sup>

Aneurysms confined to CFA, can easily be managed by open surgery with an interposition graft from the external iliac artery to the distal CFA.<sup>11</sup> Besides, those CFA aneurysms that extend into SFA or DFA can be managed with a bifurcated graft or end-to-end anastomosis to the DFA with reimplantation of the SFA or vice-versa.<sup>11</sup> Despite advancements in endovascular treatments, CFA is still a major challenge in this field because of its short length, its close proximity to the hip joint and inguinal ligament, the need for sealing zones and preserving perfusion to the SFA and PFA.<sup>11</sup> Underlying relapsing remitting inflammation in the vessel wall also adds to the complexity of this case. In view of the above, open surgery was a better option for the management of the present case. Moreover, sacrifice of DFA has multiple challenges and puts the patient in the risk of



profound ischemia to the lower limb. DFA could also be saved by open surgery. However, in our patient, the open surgical repair could not be an option since the anesthesiology service estimated a high risk for general anesthesia and open surgery according to the patient's cardiopulmonary insufficiency.

Our patient was in the active phase of the BD on admission. It has been demonstrated that the risk of post-operative complications, including wound dehiscence and aneurysm formation, is higher when the invasive procedure is done in the active phase of the disease.<sup>7,12</sup> As no signs of active bleeding or limb ischemia were evident, the managing team chose to administer immunosuppressives to reduce the disease activity before any invasive procedure was attempted. There are only a few reports on treating ruptured PAA secondary to BD in the literature.<sup>13-16</sup> All of these cases have undergone emergent ET with the exception of 1 case in which a ruptured popliteal artery aneurysm was diagnosed.<sup>14</sup> This case was also treated with immunosuppressives and closely followed for 5 days prior to ET.

For follow-up, we performed doppler examination every 6 months following the procedure, and the patient was also regularly visited by the rheumatology service. Medical treatments including corticosteroids, cyclosporin, and antiplatelets were continued following the procedure.

A combination of medical and endovascular treatment appeared as an ideal approach to our patient as it provided successful repair of the pseudoaneurysm without any evident complication. Nevertheless, evidence is very limited in this regard and a single case report cannot propose any recommendation. Additional research is needed to determine the best treatment option for these patients.

## Compliance with Ethical Standards

This study was approved by the ethics committee of Tabriz University of Medical Sciences (Approval Number: IR.TBZ-MED.REC.1400.547). Informed written consent was obtained from the patient included in the study to publish the case, medical images, and the laboratory data.

## Author Contributions

All the Authors Contributed Substantially in all Steps of This Case Report Study and Fulfill the Authorship Criteria

## Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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