

# Ruptured true aneurysm of the superficial femoral artery leading to Behçet disease diagnosis

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## ABSTRACT

Femoral artery aneurysms are rare, and aneurysms of the superficial femoral artery (SFA) are even rarer, with ruptured true aneurysms of the SFA the rarest. In the present report, we have described the case of a young patient whose SFA had clinical findings suggestive of an aneurysm rupture, which resulted in the diagnosis of Behçet disease, in accordance with the clinical features of the disease. The patient underwent standard treatment, with aneurysmectomy and interposition of a synthetic graft. (*J Vasc Surg Cases Innov Tech* 2022;8:473-6.)

**Keywords:** Behçet disease; Ruptured aneurysm; Superficial femoral artery; True femoral aneurysm

Isolated, true aneurysms of the superficial femoral artery (SFA) are very rare, occurring in 1% of all femoral artery aneurysms and 0.5% of peripheral aneurysms.<sup>1,2</sup> They will usually be found incidentally, except for urgent cases, such as peripheral ischemia due to embolization or thrombosis, a painful and pulsating mass, or aneurysm rupture, which will require immediate surgical treatment. Many factors are involved in the development of SFA aneurysms, especially in young patients. However, in the case we have described, the clinical signs and symptoms and physical and imaging findings led to the conclusion that the patient had Behçet disease (BD).<sup>3</sup> We have presented the case of a ruptured SFA aneurysm in a young Arabian male patient that resulted in the diagnosis of BD.

## CASE REPORT

A 35-year-old Arabian man sought emergency hospital treatment for sudden pain and progressive right thigh swelling of ~1 month's duration. The patient's medical history did not include any trauma, surgery, or chronic disease. He did not take any drugs or medicine, although he was a smoker. During the vascular consultation, the lower limb's femoral pulses were normal and the popliteal and distal pulses were subdued, but present. A large pulsating mass was found in the middle of the right thigh (*Fig 1, A*). Emergency color Doppler ultrasound of the artery revealed a mass, with a greatest diameter of

9.0 cm in the mid-third of the SFA and turbulent interior blood flow, suggesting a ruptured aneurysm of the artery. Computed tomography arteriography showed that the abdominal and iliac arteries were patent with no obstructions. Also, the mid-segment of the right SFA was aneurysmatic with a diameter of 8.6 cm and was compressed by the adjacent hemorrhagic mass. The distal superficial femoral, popliteal, and distal arteries had collateral flow. Additionally, occlusion of the SFA of the left thigh was revealed, without presenting symptoms (*Fig 2, A and B*). The patient was rushed to the operating room, and exploratory surgery of the right lower limb was performed. A large volume hematoma related to the ruptured aneurysm of the right SFA was present (*Fig 3, A*). The aneurysm was resected, and the SFA segment was reconstructed by interposition of a polytetrafluoroethylene synthetic graft with end-to-end proximal and distal anastomoses (*Fig 3, B*), without complications. Fragments of tissue were removed during surgery and sent for microscopic examination and bacteriologic tests. However, no germ growth was detected.

During his hospitalization, the physical examination revealed multiple genital ulcers (*Fig 1, B*) and oral ulcers (*Fig 1, C and D*). The patient underwent echocardiography during the postoperative period, with normal results. The laboratory tests at admission disclosed high levels of the acute phase reactants C-reactive protein (153.3 mg/L) and erythrocyte sedimentation rate (38 mm/h). The tests for human immunodeficiency virus, hepatotropic viruses, treponema, syphilis, cryoglobulins, rheumatoid factor, complement, and antinuclear, antineutrophil cytoplasmic, and antiphospholipid antibodies were negative. The ELISA proteinase 3, ELISA myeloperoxidase, cytoplasmic antineutrophil cytoplasmic antibody indirect immunofluorescence, perinuclear antineutrophil cytoplasmic antibody indirect immunofluorescence, rapid plasma reagin/Venereal Disease Research Laboratory, treponema pallidum hemagglutination tests were also negative. Chest computed tomography was performed, which revealed no aneurysmal findings and no involvement of the pulmonary artery.

However, because of high suspicion of BD, the patient was prescribed cortisone and colchicine by a rheumatologist. An

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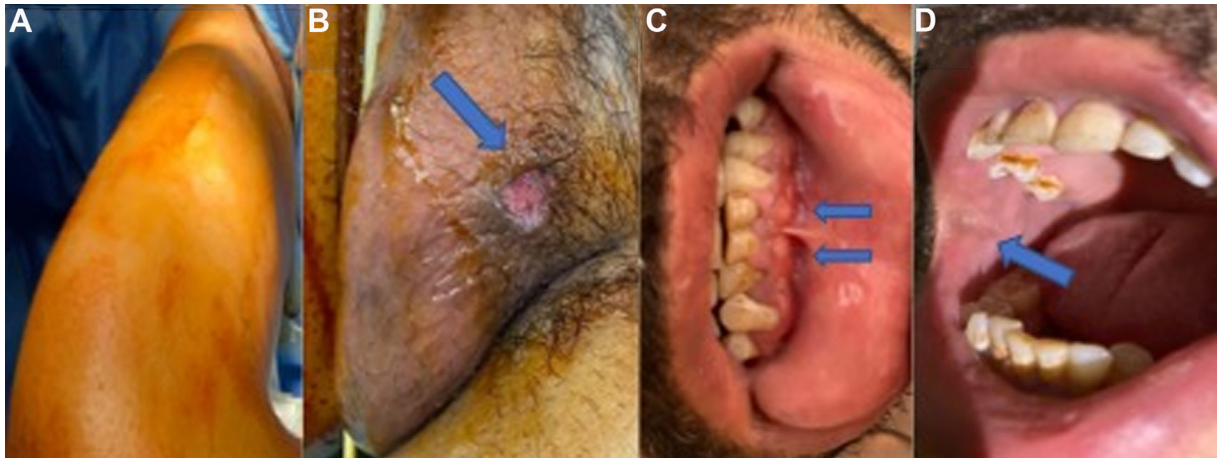
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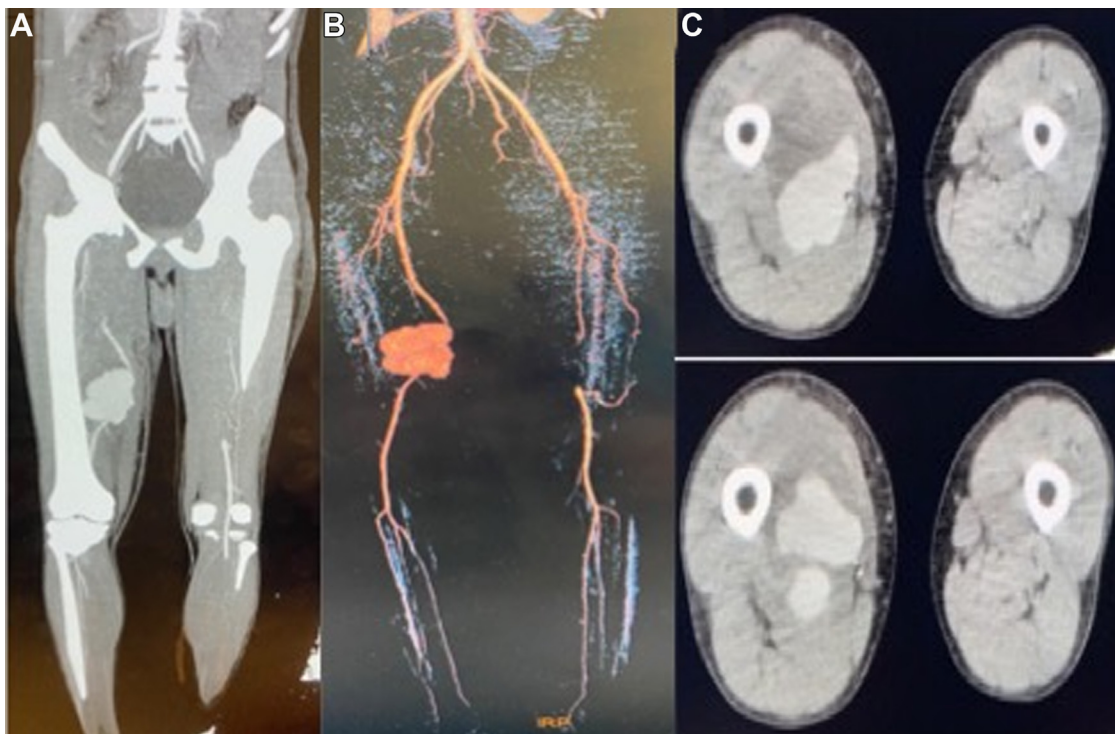
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**Fig 1.** Pulsating mass of the right thigh (A), genital ulcers (B; arrow) and oral aphthous ulcers (C, D; arrows).



**Fig 2.** Computed tomography angiogram of the lower limbs illustrating the ruptured right superficial femoral artery (SFA) aneurysm and occlusion of the left SFA, including coronal maximum intensity projection (A), three-dimensional reconstruction (B), and cross-section view (C).

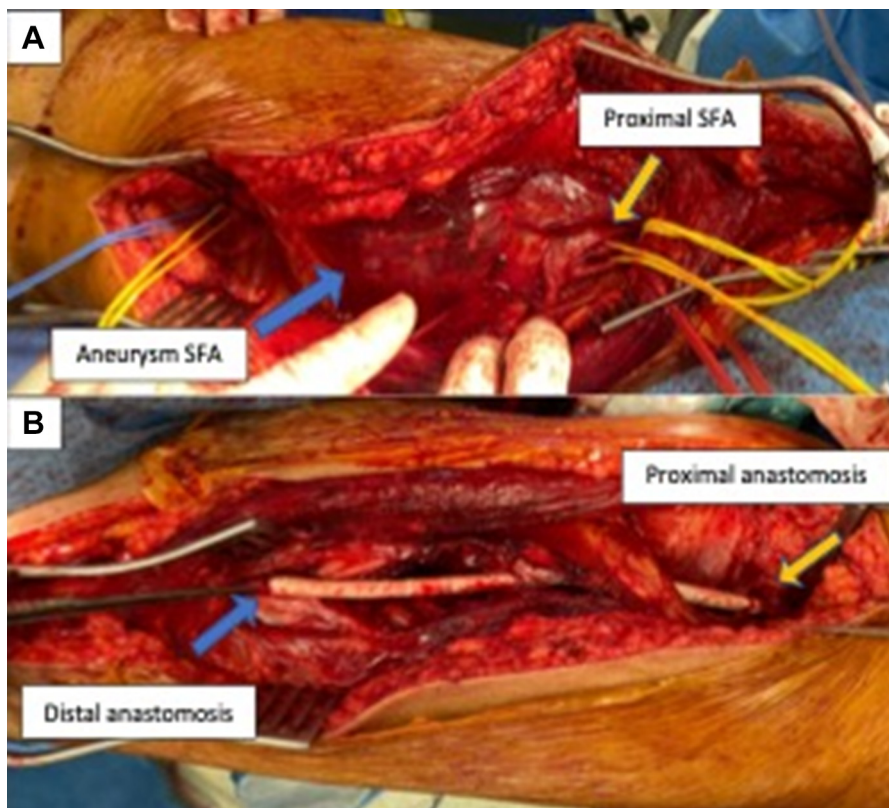
ophthalmology evaluation revealed no diplopia, swelling of both eye optic nerves, and a few scattered hemorrhages in the right eye. No pathologic findings from magnetic resonance imaging of the brain and orbit were seen. Visual field testing was not performed because of poor communication and the lack of a common language. A neurologic evaluation was performed, without pathologic evidence of neurologic BD found.

The aneurysm histologic examination revealed that the fragments had come from a large vessel wall, which had had extensive ischemic necrosis accompanied by diffuse bleeding

perspiration on presentation. The patient remained in the hospital for 15 days to complete the clinical and laboratory examinations in the context of a BD investigation. The patient provided written informed consent for the report of his case details and imaging studies.

## DISCUSSION

Aneurysms of the SFA are very rare, occurring in 1% of all femoral artery aneurysms and 0.5% of peripheral aneurysms, most often located in the distal third of the artery.



**Fig 3.** **A,** Ruptured aneurysm of superficial femoral artery (SFA; blue arrow). **B,** Aneurysmectomy and revascularization with a synthetic graft in an end-to-end anastomosis, proximal anastomosis (yellow arrow), and distal anastomosis (blue arrow).

The complications from aneurysms of the SFA have been less common than those of popliteal artery aneurysms owing to their location within Hunter's canal. They will usually be diagnosed incidentally until complications have developed.<sup>4</sup>

To the best of our knowledge, only a few dozen cases of true SFA aneurysms have been reported, with most studies reporting true atherosclerotic aneurysms. Only a few cases have referred to true SFA aneurysms resulting from BD. The highest incidence has been found in Asia and the Mediterranean countries.<sup>5</sup> Our patient was from Egypt and had presented with the characteristic features of oral and genital aphthous ulcers, eye lesions, and vascular manifestations. The 2014 Revised International Criteria for Behçet's disease were used.<sup>6</sup> The score for our patient was 7 ( $\geq 4$ ), indicating a definitive diagnosis of BD.<sup>6</sup>

Arterial involvement is rare, complicating the clinical picture and causing potentially fatal complications. The surgical treatment of a BD aneurysm can be challenging for vascular surgeons because of technical difficulties.<sup>7,8</sup> Our patient was urgently admitted to our department because of rupture of a true SFA aneurysm in the right thigh with concomitant occlusion of the left SFA.

The treatment options for patients with arterial aneurysms include surgical grafting bypass with either a vein or synthetic materials and endovascular stent implantation combined with intravenous immunosuppressant agents.<sup>7,9,10</sup> For our patient, we used a synthetic graft owing to the urgency and because the vein was deemed unsuitable owing to the venous insufficiency of the lower extremities.

The main advantage of endovascular intervention is the lower mortality rates of  $\leq 0.6\%$  to  $3.5\%$ ; however, its use is usually limited.<sup>9</sup> For our patient, we preferred open intervention with a synthetic graft bypass because of the long length and diameter of the aneurysm and the presence of a large hematoma, which had caused arterial pressure and reduced peripheral flow. In addition, our patient's young age required avoidance of an endovascular approach for SFA aneurysm treatment.

Our patient responded well after the surgical procedure, without any complications, including no infection, recurrence of aneurysmal lesions, graft restenosis, and/or thrombosis. However, the patient was lost to follow-up 5 months after the vascular surgery. As the findings for our patient have illustrated, BD poses a diagnostic challenge, especially when recognized for the first time

through unique clinical, laboratory, imaging, and histologic features.

## CONCLUSIONS

Our case report is rare because we have described the uncommon case of a patient with a true ruptured aneurysm of the SFA due to BD. Clinical suspicion should always be raised when evaluating a pulsating mass in the thigh. Treatment with open surgery seems to be a good choice, especially for younger patients. The diagnosis should be made as soon as possible to allow immunosuppressive treatment to begin in these patients, who must be monitored regularly to avoid disease complications.

## REFERENCES

1. Rutherford RB. Superficial femoral artery aneurysm. In: Rutherford RB, editor. *Vascular Surgery*. WB Saunders; 2005. p. 1538-9.
2. Oliveira FA, Oliveira HO. Aneurisma de artéria femoral superficial roto: relato de caso e revisão de literatura. *J Vasc Bras* 2009;8:285-8.
3. Vilariño-Rico J, Vidal-Insua JJ, Cachaldora JA, Segura-Iglesias RJ. Aneurismas verdaderos de la femoral superficial: a propósito de un caso. *Angiol (Barcelona)* 2008;60:149-54.
4. Dobritz M, Lang W, Fellner FA. MR diagnosis of superficial femoral artery aneurysm, primarily misdiagnosed as soft tissue tumor: a case report. *Magn Res Imaging* 2003;21:687-90.
5. Behçet H. Über rezidivierende Aphthose, durch ein Virus verursachte Geschwüre am Mund, am Auge, und an den Genitalien. *Dermatol Wochenschr* 1937;36:1152-7.
6. International Team for the Revision of the International Criteria for Behçet's Disease (ITR-ICBD). The International Criteria for Behçet's Disease (ICBD): a collaborative study of 27 countries on the sensitivity and specificity of the new criteria. *J Eur Acad Dermatol Venereol* 2014;28:338-47.
7. Merashli M, Eid RE, Uthman I. A review of current management of vasculo-Behçet's. *Curr Opin Rheumatol* 2018;30:50-6.
8. Hussein MK, Ahmed AS. Outcomes of segmental femoral artery pseudoaneurysm in patients with Behçet's disease: a single center's experience. *J Vasc Bras* 2019;18:e20180132.
9. Sallustro M, Faggioli C, Ancetti S, Gallitto E, Vento V, Pini R, et al. Endovascular treatment of a ruptured superficial femoral artery aneurysm in Behçet's disease: case report and literature review. *Ann Vasc Surg* 2020;65:287.e1-6.
10. Yang SS, Park KM, Park YJ, Kim YW, Do YS, Park HS, et al. Peripheral arterial involvement in Behçet's disease: an analysis of the results from a Korean referral center. *Rheumatol Int* 2013;33:2101-8.

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