

# Management of tibioperoneal trunk aneurysm in a patient with Behçet disease

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## ABSTRACT

Only a few cases of infrapopliteal aneurysms are reported in the literature. These are commonly associated with trauma, infection, and iatrogenic injuries and mostly present as pseudoaneurysms. We report the case of a 44-year-old man with Behçet disease and an 8-cm tibioperoneal trunk aneurysm and discuss the management options of these aneurysms. (*J Vasc Surg Cases and Innovative Techniques* 2018;4:15-8.)

Behçet disease is an autoimmune disease characterized by systemic vasculitis and a triple complex of symptoms: recurrent oral aphthous stomatitis, uveitis, and genital ulcers. Inhabitants of Turkey and Far Eastern Asian countries have the highest prevalence of the disease.<sup>1</sup> Although an arterial lesion secondary to Behçet disease is a rare manifestation, an increased frequency of recurrence and life-threatening complications including ischemic bowel perforations, sudden rupture of large arterial or aortic aneurysms, and cerebrovascular accidents bring challenges to providing treatment. Immunosuppressive therapy (eg, oral prednisolone) is administered to reduce the risk of such outcomes.<sup>2</sup> Medically treated aneurysmal lesions respond poorly, and in most cases, surgery is mandatory.<sup>3</sup> However, surgical intervention is challenging in these patients compared with patients with normal vascular integrity. The intense local inflammation may increase complications. In addition, arterial aneurysms in patients with Behçet disease are at a high risk of rupture, and patients are more likely to have multiple lesions at several sites. The aim of this report was to highlight the management of a tibioperoneal trunk (TPT) aneurysm in a patient with Behçet disease. The patient has consented to this presentation.

## CASE REPORT

**History.** A 44-year-old man presented to the vascular outpatient clinic for evaluation of an infrarenal abdominal aortic aneurysm and a right popliteal aneurysm. The patient was

hypertensive and had a history of Behçet disease characterized by orogenital ulcers and posterior uveitis with left eye blindness. The patient has been receiving steroid therapy since his diagnosis 14 years ago and underwent a left 8 × 5-cm popliteal aneurysm repair 5 months before presentation in an outpatient setting outside the United States. He is a current every-day smoker of 100 pack-years.

On physical examination, the patient had normal vital signs, a weight of 80 kg, a height of 176 cm, and a BMI of 25.8 kg/m<sup>2</sup>. Review of systems was positive for blurred vision and diffuse rash. Physical examination of both legs showed no evidence of infection, cyanosis, or edema. Abdominal examination showed no palpable or pulsating masses. Lower extremity pulses were 2+ and symmetric except for a prominent 4+ right popliteal artery pulsation. Laboratory test results were within normal limits.

**Imaging.** Lower extremity Doppler ultrasound examination showed a right below-knee popliteal artery aneurysm measuring 4.08 × 3.74 × 3.91 cm and a patent left above-knee popliteal to proximal posterior tibial artery bypass with normal graft velocity. It appeared that the left anterior tibial artery was ligated, and a collateral flow filled back into the peroneal artery. Ankle-brachial index was 1.2 on the right side and 1.12 on the left side. Digital pressure was 139 mm Hg and 104 mm Hg, respectively. Duplex ultrasound of the abdominal aorta showed a 3.4-cm saccular infrarenal aneurysm. Bilateral lower extremity and aortic angiography was performed and showed a normal right popliteal artery with a large 7- × 8-cm saccular TPT aneurysm (*Fig 1*). A 3- to 4-cm saccular aneurysm in the mid to distal aorta was seen on the aortic angiogram.

**Operation.** The risks and benefits of the TPT aneurysm repair were discussed with the patient. These included the high risk of aneurysm recurrence, given his vasculitis condition, and the importance of lifetime anticoagulation to decrease the risk of thrombosis and graft occlusion. He consented to the open repair.

The patient was intubated and placed in the prone position. We made a gentle S-shaped incision extending from above to below the knee and identified the lesser saphenous vein, which was followed to the junction with the popliteal vein proximally and then to about midcalf. All tributaries were doubly ligated. The popliteal artery was identified behind the knee and was dissected free, with vessel loops placed around it. We followed

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**Fig 1.** Preoperative angiographic image of the right lower extremity showing a large 7- × 8-cm saccular tibioperoneal trunk (TPT) aneurysm.

it down to the large TPT aneurysm extending 8 cm into the deep calf muscle. The dissection was difficult and tedious and took >3 hours. The aneurysm was completely freed up from all the surrounding tissue with no bleeding (Fig 2, A). The posterior tibial and peroneal arteries were identified and controlled with the vessel loop. The patient was given 5000 units of heparin intravenously. After the sac was opened, there was backbleeding from several small branches, which were suture ligated with a figure-of-8 5-0 Prolene from within the aneurysm. The vein was transected proximally and distally and dilated gently with heparinized saline to approximately 5 mm in diameter. The vein was inverted, and two valves were identified and excised. The proximal end of the vein was sewn to the popliteal artery below the knee. We were able to include the anterior tibial artery in the back wall of the anastomosis. The vein was cut to the proper length and was sutured to the posterior tibial artery distally (Fig 2, B). We did transect the aneurysm distally and oversewed the TPT with two layers of nylon. At the end, there were palpable dorsalis pedis and posterior tibial pulses in the foot, confirmed with triphasic Doppler signals that dissipated on clamping and were augmented when the bypass was released. The patient was stable hemodynamically throughout the case and was transferred in stable condition to the recovery room.

**Discharge and follow-up.** The patient was discharged home after 3 days with an uneventful postoperative course. Follow up duplex ultrasound at 4 weeks, 12 weeks, 6 months, and 1 year showed widely patent right lower extremity bypass (Fig 3). The ankle-brachial index and toe pressure were normal bilaterally. The abdominal aortic aneurysm was stable in size.

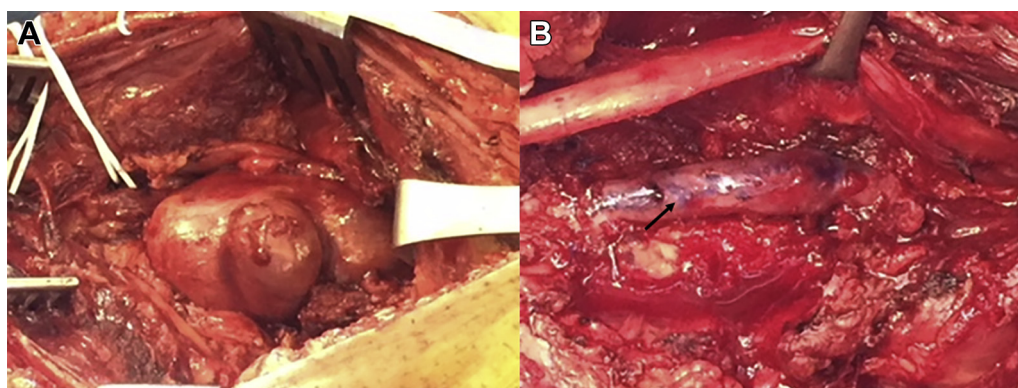
## DISCUSSION

Arterial complications occur in 25% to 30% of patients with Behçet disease, with aneurysms occurring anywhere throughout the body, either synchronously or metachronously. Aneurysmal degeneration in this disease is probably due to obliterative endarteritis of the vasa vasorum supplying the medium-sized and large vessels.<sup>4</sup> These are more preferably treated with endovascular than with open repair.<sup>5-8</sup> Endovascular procedures are effective and safe, with acceptable complication rates and good durability.<sup>2</sup>

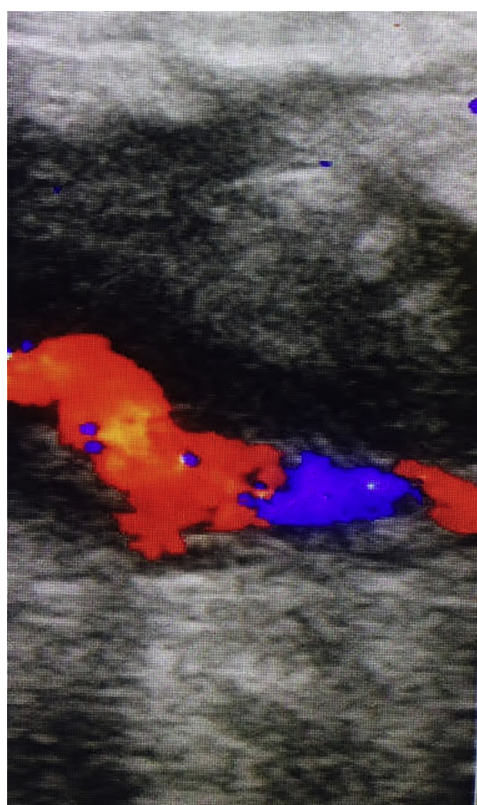
In this report, we present the case of a male patient with Behçet disease who had undergone prior successful left TPT aneurysm resection and repair with above-knee popliteal to posterior tibial bypass. The patient presented with a large right TPT aneurysm reaching 8 cm in diameter. There was no proximal or distal neck at which to place the covered stent, and the aneurysm was very large with no neck to coil. As a result, the patient was not a candidate for the endovascular approach.

Tibioperoneal artery aneurysms are rare and are mostly due to trauma, infection, and iatrogenic injuries.<sup>9</sup> After careful literature search, we identified eight cases of patients with TPT aneurysms,<sup>4,9-14</sup> only one of which was secondary to Behçet disease.<sup>4</sup> The aneurysm was 6.6 cm in diameter and was resected, followed by a below-knee popliteal to distal posterior tibial artery bypass using reversed ipsilateral great saphenous vein graft. Two of the other reported TPT aneurysms were due to atherosclerosis,<sup>13,14</sup> and the remaining five were of unspecified etiology.<sup>9-13</sup> On the other hand, in 2011, Rico et al<sup>15</sup> reported a case of a 41-year-old man with Behçet disease who presented with a ruptured tibioperoneal pseudoaneurysm. The patient underwent emergent endovascular embolization with three 5-mm metallic coils.

There is no consensus as to whether to perform bypass surgery or endovascular stent placement for aneurysm repair secondary to Behçet disease.<sup>16</sup> The endovascular approach is preferred in Behçet disease patients with suitable anatomy (usually aortic aneurysms) because of its relatively lower morbidity and mortality rates compared with open surgical repair. However, in peripheral aneurysms secondary to Behçet disease, the stent grafts have a greater risk of complications, such as thrombosis and development of a pseudoaneurysm at the site of the repair.<sup>3,17</sup> On the other hand, possible procedure-related complications after open aneurysm excision and bypass include acute graft occlusion, long-term restenosis, aneurysm recurrence, and fistula



**Fig 2.** Intraoperative pictures of the (A) tibioperoneal trunk (TPT) aneurysm and (B) lower extremity bypass. The arrow in (B) shows the vein conduit. The trocar is holding the tibial nerve.



**Fig 3.** Follow-up duplex ultrasound examination of the right lower extremity. The right lower extremity bypass is widely patent at 1 year postoperatively.

formation at the anastomotic site.<sup>4</sup> The establishment of remission before the surgical intervention has been shown to decrease postoperative complications. Immunosuppressive therapy, such as oral prednisone, azathioprine, cyclosporine, and others, is given before and after surgery to prevent early and late postoperative complications.<sup>18</sup> Some authors recommend using corticosteroids combined with immunosuppressants for at least 2 years postoperatively to decrease the risk of recurrent aneurysms.<sup>16,19</sup>

## CONCLUSIONS

Tibioperoneal aneurysms are a rare manifestation of Behçet disease. Cases should be evaluated on an individual basis, and management depends mainly on the location, anatomy, and size of the aneurysm. Close follow-up is recommended to monitor for disease progression and recurrence as well as to detect postoperative complications.

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