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Spontaneous Pseudoaneurysm of the Superficial Femoral Artery in Behcet's Disease—Endovascular Stent-Graft Treatment Combined with Percutaneous Drainage: A Case Report

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ABSTRACT

Behcet's disease is a rare multisystemic chronic autoimmune disorder characterized by a classic triad of urogenital ulcerations, chronic eye inflammation, and skin lesions. We report on a case of a spontaneous pseudoaneurysm of the superficial femoral artery caused by Behcet's disease that was treated with an endovascular stent-graft followed by percutaneous drainage. We emphasize the use of percutaneous drainage of the pseudoaneurysm to decrease compression on the stent-graft and native vessel.

INTRODUCTION

Behcet's disease is encountered throughout the world, especially in the Mediterranean, Middle East, and Eastern Asia. Clinically, it is most commonly seen as systemic vasculitis (venous thromboembolism and arterial pseudoaneurysm), which characterizes almost 30% of the cases. Arterial complications account for 10% to 15% of vascular complications and consist mainly of aneurysms [Park 2001].

CASE REPORT

A 41-year-old man presented a growing mass on his right thigh region. Ten years before, he had been hospitalized and treated for Behcet's disease at another hospital. There was severe pain in the right lower limb. The mass was pulsatile, and a Doppler examination revealed a pseudoaneurysm with dimensions of 4 × 5 cm. There was also fistulization into the femoral vein. A Doppler ultrasound scan of the femoral arteries found evidence of a pseudoaneurysm in the right superficial femoral artery (SFA). There was a small amount of thrombus in the aneurysm. Digital subtraction angiography confirmed a large SFA pseudoaneurysm (Figure 1). The neck of the aneurysm was 3 mm long. There was no evidence of associated vascular pathology and only minimal thrombus formation at the level of the aneurysmal wall, which placed the patient at a relatively low risk for embolic complications during an endovascular procedure. Because of the aneurysm, the most appropriate therapy was determined to be endovascular placement of a covered stent. Under local anesthesia and full heparinization, an 18 F, 11-cm sheath was inserted percutaneously in the right femoral artery by an ipsilateral-antegrade route. An angiography was obtained via the sheath and a 0.018 guide wire was crossed from the aneurysm. A left anterior oblique 45° image was selected as a working projection on which the aneurysm neck was well demonstrated. A 28-mm long balloon-expandable stent (4 mm to 9 mm diameter range) with an internal polytetrafluoroethylene covering (Jostent Peripheral Stent Graft; Abbott, Abbott Park, IL, USA) was manually crimped on a 6 × 40-mm angioplasty balloon catheter (Fox plus PTA Catheter; Abbott). The PTA balloon catheter was guided over the wire through the sheath and positioned at the level of the neck. The balloon inflated to 14 atmospheres to create the 6.5-mm diameter area needed to deploy the stent-graft. A control angiography showed a complete closure of the pseudoaneurysm. An 18 G needle was inserted in the pseudoaneurysm far enough away from the stent-graft to avoid touching it (Figure 2). When the stylet was pulled out, a contrast media gushed out (flowing with pressure), and after 100 mL of drainage, another 100 mL of bloody fluid flowed through manual compression. The mass lesion became soft and pressure on the native vessel and stent graft returned to normal. The patient felt relief from the pressure and pain, experienced no complications, and was discharged one day later on an antiplatelet regimen consisting of 75 mg of clopidogrel for 6 months and aspirin (100 mg daily) indefinitely.

DISCUSSION

Behcet's disease is a rare, chronic, autoimmune, multisystem disorder that causes inflammation of blood vessels (vasculitis)

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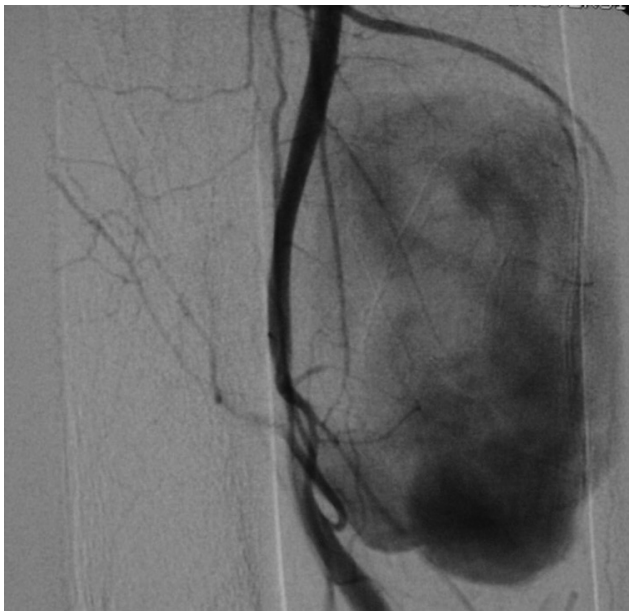

Figure 1. DSA revealed a large superficial femoral artery pseudoaneurysm.

anywhere in the body. The predominant vascular lesions in Behcet's disease are arterial and venous occlusions and aneurysm formation. Venous lesions constitute the majority of vascular pathologies (88%), while arterial involvement constitutes the rest (12%) [Park 2001]. The pathogenesis of the aneurysm formation or rupture seems to be vasculitis resulting in obliterative endarteritis of the vasa vasorum [Matsumoto 1991]. A pseudoaneurysm is a dilatation of an artery with arterial disruption of one or more layers of the wall, and can occur through puncture as a complication of percutaneous arterial catheterization, rather than expansion of all layers of the wall. Mainly pulmonary, femoral, iliac, aortic, and popliteal aneurysm formations are seen. Pseudoaneurysms in the arterial system are seen most commonly on the femoral artery. Pseodoaneurysms of the SFA are usually traumatic or iatrogenic in origin. Adventitial thickening, fibrosis and perivascular lymphocytic infiltration, a decrease in elastic and muscular fibers in the media layer, and an increase in smooth muscle and fibroblastic cells in the intimal layer are the main pathologic findings in an aneurysmal wall. Obliterative endarteritis of the vasa vasorum is thought to be the cause of an aneurysmal dilatation.

Fragility and weakness of the vascular wall due to arteritis are generally responsible for postoperative pseudoaneurysm formation at the anastomotic site in these cases. The pathology in Behcet's disease is not similar to that found in Ehlers-Danlos syndrome. During conventional operations in Behcet patients, the arterial walls have enough strength to hold sutures, but in the later period complications occur [Alhan 1999; Akpolat 2000; Kutlu 2002]. Although rupture of an arterial aneurysm is a rare event, it is the leading cause of death in patients with Behcet's disease. The pathogenesis of the aneurysm formation or rupture seems to be vasculitis resulting in obliterative endarteritis of the vasa vasorum. To prevent its rupture, a surgical procedure can be performed to resect the aneurysm and replace it with a graft. However, recurrence of a pseudoaneurysm after its resection, especially at the site of surgical repair, occurs in approximately 50% of cases and the fragility of the vascular wall may play a major role in this situation. Conventional treatment by resection and placement of a prosthetic or autogenous vein graft is an effective but technically demanding surgery. Endovascular stent-graft implantation is a minimally invasive procedure compared to open surgery and can be performed successfully in patients with a pseudoaneurysm. Since the early 1980s, endovascular stents and, more recently, stent-grafts have become the preferred treatment for occlusive lesions, dissections, and vascular malformations in most circulatory beds. The success of using percutaneous endovascular treatment for peripheral aneurysms has recently been reported [Henry 2000; Casana 2003]. The effectiveness of endovascular stent-grafting for aortic and arterial aneurysms in patients with Behcet's disease has been also demonstrated, and the stent-graft may represent a responsible alternative to open surgery because of the high recurrence rate after such surgery [Okada 1997; Sasaki 1998; Henry 2000; Park 2001; Casana 2003].

When evaluating a candidate for stent-grafting, morphological assessment of the lesion by color-flow duplex scanning, computed tomographic angiography, and digital subtraction angiography provide important data for selecting the appropriate device. Most saccular aneurysms, even those with a thrombus, may be considered appropriate for stent-grafting. However, the presence of an unstable thrombus in the parent artery is a contraindication to endovascular maneuvers. We also consider intraoperative heparinization and antiplatelet therapy mandatory to prevent clot formation within the device and to minimize embolic risk even when

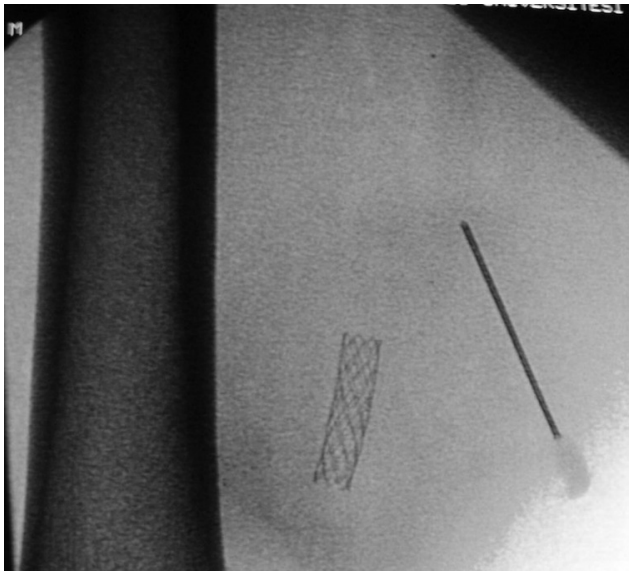

Figure 2. Post-intervention direct imaging shows the stent and the needle.

there is no thrombus at the level of the lesion. We routinely prescribe antiplatelet agents for months after carotid stenting, but long-term anticoagulation does not appear to be necessary [White 1995]. We used an 18 F sheath when we were conducting this study; however, we recommend 18 F or higher calibers to provide sufficient working space. Moreover, high-quality fluoroscopic imaging with multiple projections was mandatory to achieve an angiographic assessment of the stent-graft placement.

In past cases such as this, the stent mesh impeded flow into the aneurysmal sac, thus inducing thrombosis, or relocated the intimal flap to occlude the aneurysm. However, it was difficult to exclude flow into the wide-necked aneurysms with an uncovered stent, so coil embolization of the sac through the stent was often required. Today, wide-necked saccular aneurysms or pseudoaneurysms and lengthy tears in the arterial wall can more appropriately be treated with a covered stent, leading to immediate and definitive reconstruction of the arterial wall [Ruebben 1997].

The main advantages of endovascular treatment are lower mortality levels, ranging from 0.6% to 3.5%, even in high-risk groups, and a high success rate (97%). Percutaneous stent-graft insertion offers many advantages, including a short procedure time, less invasiveness, immediate isolation of the aneurysm and bleeding site from circulation without the need for general anesthesia, minimal blood loss, and reduced length of hospital stay, morbidity, and mortality [Kasirajan 2001]. These advantages are especially relevant in those patients facing high risk in surgery because of advanced cerebrovascular, cardiovascular, or other systemic disease. The overall complication rate is low, but reported complications include distal migration of the stent, residual leak, late growth of the aneurysmal cavity, distal embolization, infection, and contralateral venous thrombosis in patients with an iliac artery aneurysm [Razavi 1995; Christensen 1996].

It is also important to remember that endovascular treatment with a stent-graft may not relieve the compressive symptoms caused by the aneurysm. The fate of a thrombosed aneurysm is not clearly understood. Therefore, in patients with symptoms caused by compression from the aneurysmal sac, this treatment method may not be indicated unless an associated medical disease precludes surgery. On the other hand, surgery for pseudoaneurysms may produce an aneurysmal hematoma that results in quick pain relief but pressure on the vessels. Removal of an aneurysm by percutaneous intervention decreases the pressure on the stent-graft and native vessel. This procedure also lowers to a minimum the risk of microorganism contamination during surgery. Furthermore, it eliminates the creation of a space that would remain following the surgical excision of the aneurysm and encourage development of a new hematoma. If puncture devices with the possible smallest diameters are used during percutaneous drainage, possible fistula formation between the aneurysm sac and the surface of the skin can be prevented. Since the caliber 18 puncture needle, which we used in our case, is quite a small-gauge material, no fistula formation was observed.

In conclusion, implantation of a stent-graft for the treatment of pseudoaneurysms in patients with Behcet's disease is an attractive alternative technique because of the shorter length of hospital stay and period of time required before returning to normal life, lack of general anesthesia, and surgical dissection. Percutaneous removal of the aneurysm sac can be done to effectively eliminate some symptoms in the early period.

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