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Case Report

Massive Hemorrhage Due to Rupture of A Right Iliac Aneurysm Revealing Behcet's Disease: A Case Report From the Festoc Center in Bamako

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Abstract

Introduction: *Behçet's disease is a chronic inflammatory disorder. It is clinically characterized by oral and genital aphthoses; skin lesions and systemic manifestations. Arterial aneurysms are defined as a loss of parallelism of the arterial walls, localized to an arterial segment, and corresponding to an increase of at least 50% of the normal arterial diameter. We report a case of Behcet's disease revealed by a ruptured aneurysm of the right primitive iliac artery.*

Observation: *This was a 44-year-old patient with no known history, referred for management of a right inguinal mass that had progressively increased in volume and had been evolving for about 1 month. On admission, the physical examination revealed conjunctival pallor, a pulsatile right inguinal mass approximately 10 cm in diameter, and a suppurating sore on the scrotum. Doppler ultrasound of the lower limbs revealed an aneurysm of the right common iliac artery. The aneurysm ruptured during the short preparation period for surgery, resulting in cataclysmic hemorrhage. This prompted manual compression and the transfusion of several packed red blood cells. During transport to the OR, the patient experienced hemorrhagic shock and altered consciousness, prompting the administration of drugs (noradrenaline) and emergency orotracheal intubation. The aneurysm was controlled upstream and downstream by approaching the right primitive iliac artery, followed by dissection and clamping after general heparinization.*

The common femoral artery was then approached and checked. Surgical exploration revealed numerous clots which were evacuated, revealing a large neck extending over 7 cm. The procedure involved reconstruction of the common femoral artery with a patch saphenous graft and excision of the necrotic tissue. The post-operative course was marked by suppuration of the surgical wound, which progressed well with appropriate antibiotics and local care.

Conclusion: *Arterial involvement in Behçet's disease manifests as arterial thrombosis or, more frequently, aneurysm. This is due to inflammation of the media, intima and vasa vasorum. Their management can be envisaged in our context.*

Introduction

Arterial aneurysms of the limbs are one of the main diseases of the blood vessels. They are defined as a loss of parallelism of the arterial walls localized to an arterial segment, and corresponding to an increase of at least 50% of normal arterial diameter. They account for 18% of all arterial aneurysms and are the most common after aortic aneurysms [1]. The etiology of limb AA can be degenerative, and these patients frequently have classic cardiovascular risk factors. In rarer cases, certain inflammatory or connective tissue diseases are the cause. These degenerative aneurysms are to be distinguished from false aneurysms of iatrogenic or post-traumatic origin.

Case report

This is a 44-year-old patient with no known medical or surgical history, referred by a family doctor for management of a right inguinal mass that had been progressively enlarging for about 1 month.

The physical examination on admission was marked by conjunctival pallor, a pulsatile right inguinal mass bleeding about 10 cm in diameter and a suppurating sore on the scrotum (Figure 1).

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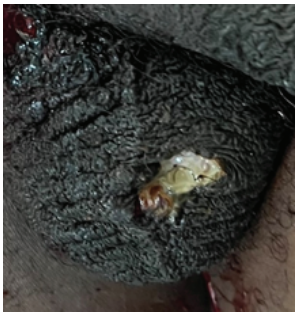

Figure 1. *Aphte génital*

Doppler ultrasound of the lower limbs revealed a right inguinal adenopathy with fistulization responsible for demodulation of arterial flow (Figures 1 & 2).

Biological tests showed microcytic anemia at 8 g/dl.

The evolution was marked by a rupture of the aneurysm during the short period of preparation for surgery, resulting in cataclysmic hemorrhage. This prompted manual compression and the transfusion of several packed red blood cells.

During transport to the operating room, the patient presented with hemorrhagic shock and altered consciousness, prompting drugging (noradrenaline) and emergency orotracheal intubation.

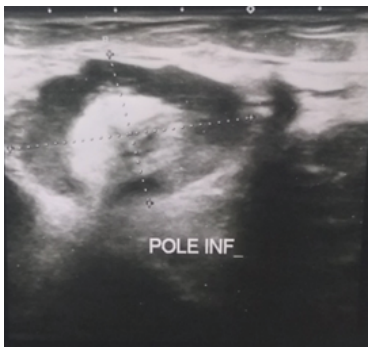

Figure 2. *Aphte génital*

Figure 3. *Aphte génital*

The aneurysm was urgently controlled upstream and downstream by approaching the right primitive iliac artery with a 4 cm incision on the line connecting the anterior superior iliac spine and the pubis, followed by dissection and clamping after general heparinization. The common femoral artery was then approached and controlled.

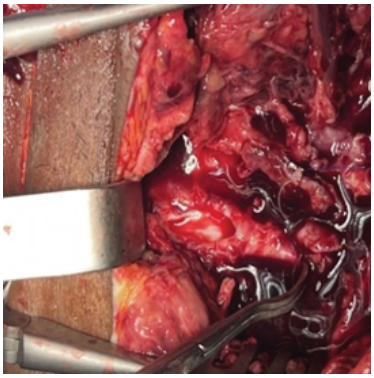

Figure 4. *Aneurysm neck*

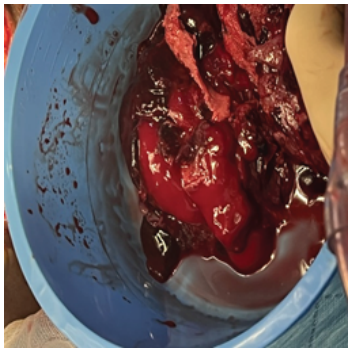

Figure 5. *Numerous clots removed*

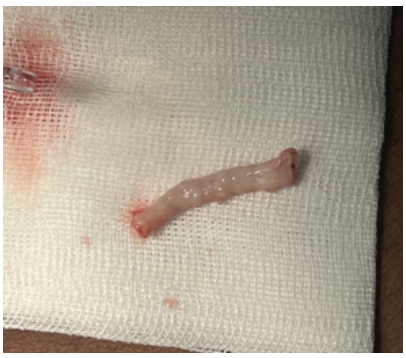

Figure 6. *Venous graft*

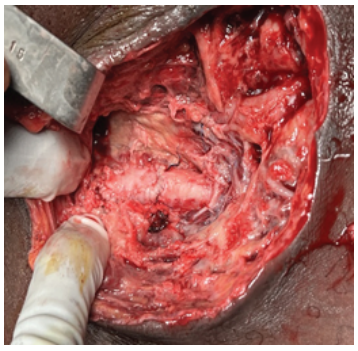

Figure 7. *Patch over the collar*

Surgical exploration, after control and opening of the aneurysm, revealed numerous clots which were evacuated, revealing a large neck extending over 7 cm (Figures 4 & 5).

The procedure involved reconstruction of the common femoral artery with a patch saphenous graft (Figures 6 & 7)

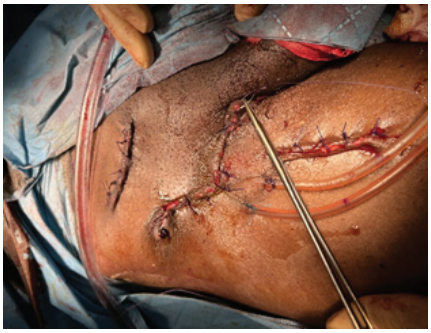

Figure 8. *Closure after cure.*

and excision of necrotic tissue.

2 redon drains were inserted prior to closure.

The post-operative course was marked by suppuration of the surgical wound, which progressed well with antibiotics and local ointments.

Discussion

Behçet's disease (BD) is a chronic systemic vasculitis of unknown etiology, characterized by recurrent oral ulcerations, genital ulcerations, ocular lesions, skin lesions and multi-organ involvement, including vessels, heart, joints, lungs, nervous and central nervous system, and gastrointestinal system [1].

The average age of onset of BD is 35 years, and both sexes are equally affected, although the severity is greater in younger men [3]. Our patient's age is above this average, but he is male.

More than a third of BD patients have vascular involvement. BD is a chronic, relapsing systemic vasculitis that shares certain characteristics with autoimmune and autoinflammatory diseases [4]. The clinical manifestations of this disease are complex and diverse. The etiology and etiopathogenesis of BD remain unclear and may be related to genetic and immunological factors, as well as microbial infection. [5-6].

Microbial infection may be a trigger for BD. Four peptides derived from the Heat 65 sequence (HSP65) have been identified by T-cell epitope mapping that specifically stimulate gd T cells in BD patients [7]. HSP65 and herpes simplex virus (HSV)-1, Streptococcus sanguis may share a common denominator, while cross-reactivity leads to an immunoreaction in individuals with hereditary susceptibility [8].

HSV1-infected patients present with clinical symptoms similar to those of BD patients, including oral aphthous ulcers, genital ulcers, eye lesions, skin lesions, gastrointestinal ulcers and arthritis.

Aneurysms associated with BD often require surgical intervention. The indications and timing of surgery depend on the anatomical position and clinical status, including whether the aneurysm is ruptured or close to rupture, and whether the disease is active or in remission [9].

In general, ruptured or near-ruptured aneurysms require prompt surgical intervention; however, the risk of postoperative complications, such as anastomotic pseudoaneurysm, aneurysm rupture, thrombosis and anastomotic infection, is high. Surgery should be avoided in patients with active vasculitis, and immunosuppressive therapy should be preferred.For the management of peripheral arterial aneurysms, which are small and asymptomatic and present a low risk of rupture, cyclophosphamide and high doses of corticosteroids are recommended [10-11].

Anti-TNF-a agents, particularly infliximab, may be a good alternative for their management in cases of cyclophosmamide intolerance [13].

Open surgery is recommended for larger vascular lesions in BD may entail a high risk of post-operative relapse, pseudoaneurysm or even aneurysm rupture, due to a large wound surface and multiple vascular anastomoses. Recently, endovascular interventional therapy, which causes less damage, fewer complications and less mortality, has recently been introduced, less damage, fewer complications and less mortality, is increasingly used for the treatment of BD-associated aneurysms [14].

Furthermore, it is suspected that BD patients tend to activate the coagulation process and/or inflammation are prone to activate the coagulation process and/or inflammatory response to induce thrombosis through dysfunction of the vessel endothelium.

However, there is as yet no consensus regarding the use of anticoagulants in patients with postoperative BD.

Conversely, it is recommended that patients with postoperative BD continue to use glucocorticoids and immunosuppressants, although the duration of treatment is difficult to determine.

However, early discontinuation of immunosuppressive therapy increased the relapse rate of BD [15].

Conclusion

Behçet's disease (BD) is a systemic inflammatory disorder of unknown etiology that progresses in relapses. Thus, the diagnosis of Behçet's disease requires the presence of recurrent oral aphthosis, associated with either genital aphthosis, uveitis, or pseudofolliculitis.

Although the treatment of MB remains highly empirical to this day, it has been well demonstrated that early and effective treatment of acute inflammatory flare-ups and prevention of relapses significantly improves the outcome of the disease.

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