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Cas clinique

Rupture of the abdominal aorta aneurysm combined with superficial femoral artery thrombosis due to Behçet's disease: a case report

Rupture d'un anévrisme de l'aorte abdominale associée à une thrombose de l'artère fémorale superficielle due à la maladie de Behçet : une observation clinique

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Résumé
La maladie de behçet est une vascularite systémique à tropisme surtout veineux. L'atteinte artérielle est peu fréquente représentée essentiellement par l'anévrisme de l'aorte abdominale. L'atteinte anévrysmale de l'aorte abdominale est trompeuse se manifestant par une symptomatologie atypique responsable d'un retard diagnostique, qui parfois peut se solder par une rupture. Ces complications peuvent être le mode de révélation de la maladie. Le patient en discussion âgé de 31 ans, était admis au service d'urgence pour des douleurs abdominales paroxystiques et un état de choc, L'enquête étiologique a conclu à un angioBehcet. Un traitement par corticoïde et immunosuppresseur a été instauré, avec succès. Devant un abdomen aigu chez un jeune patient, il faut toujours rechercher un anévrysme de l'aorte abdominale incitant à une recherche étiologique minutieuse.

Mots-clés : Bechçet, anévrisme, rupture, aorte abdominale

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Summary
Behçet's disease is a systemic vasculitis with a particular tropism for veins. Arterial involvement is rare and is mainly represented by an aneurysm of the abdominal aorta. Aneurysmal involvement of the abdominal aorta is misleading, manifesting with atypical symptoms that lead to a diagnostic delay, which rarely favors rupture. These complications can be the presenting mode of the disease. We report a case of a 31-year-old patient, who was admitted to the emergency department for paroxysmal abdominal pain and a state of shock. The etiological investigation pointed to an angio-Behçet's disease. Treatment with corticosteroids and an immunosuppressant was initiated, with good outcomes. In the case of an acute abdomen in a young patient, one should always be wary of an abdominal aortic aneurysm, which warrants a meticulous etiological investigation.

Keywords: Behçet, aneurysm, rupture, abdominal aorta

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Introduction

Behçet's disease (BD) was first defined by Hulusi Behçet, a Turkish Professor of Dermatology, in 1937 as a triad of recurrent aphthous stomatitis, genital aphthae and relapsing uveitis (1).

It is a multigenetic, chronic systemic vasculitis with an unknown origin characterized by recurrent oral ulcers, mucocutaneus disorders and ocular findings, as well as articular, vascular, neurological, pulmonary, gastrointestinal, renal and genitourinary manifestations (2).

Case presentation

A 31- year-old man with no particular medical and surgical history, was admitted to the emergency department with complaints of severe

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abdominal and back pain, progressive fatigue, and dizziness. At physical examination her abdomen was tender, and femoral artery pulses were absent. Blood pressure was 70/40 mm Hg, and cardiac auscultation was normal except for sinusal tachycardia. Chest radiographs were normal. Laboratory investigations revealed a hemoglobin level of 8 g/dL and hematocrit of 21%. Abdominal ultrasonography and CT-angiography revealed an abdominal aortic saccular aneurysm measuring (Antero-Posterior x Transverse x Cranio-Caudal) 63 × 50 ×07 mm, complicated by a hematoma dissecting the aortic wall with signs of rupture retroperitoneal. Associated vascular enhancement of the aortic wall with absence of parietal calcifications suggests first a vascular or even infectious origin; the athermanous origin is not in the foreground (figure 1). The patient was taken to the operating theater urgently. After median laparotomy, the large hematoma was seen in the retroperitoneal field. Abdominal aorta reconstruction was performed with a straight Polyester (Dacron®) prosthesis, below the aorta and the iliac artery bifurcation.

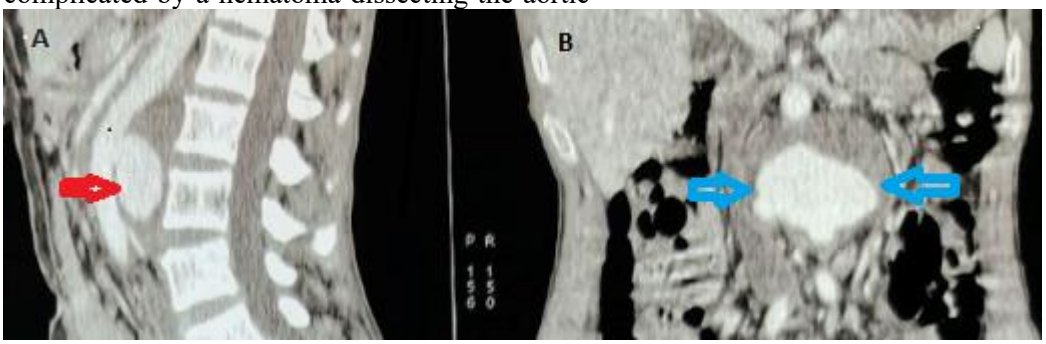

Figure 1. sagittal (A) and axial (B) section on preoperative computed tomography scan showing a Large pre-vertebral saccular aneurysm (red arrow) of the renal aorta with intra- and peri-aortic hematoma (blue arrow)

The patient had a favorable postoperative course with gradual improvement of his back pain.

A biological assessment with an etiological aim was requested, objectivean inflammatory fluid with a white blood cell count of 1660 cells/mm^3 made of 70% of neutrophils, and an erythrocyte sedimentation rate at 96 mm/h and a C-reactive protein at 100 mg/l. Gram stain and bacterial culture were negative. Sputum testing for tuberculosis was negative. Chest X-ray was normal. Radiography of the spine, sacroiliac joints, and knees showed no sign of spondyloarthritis. Syphilis testing in the blood and joint fluid was negative. Serology testing for hepatitis B virus, hepatitis C virus, and HIV was negative. Rheumatoid factor and anti-CCP, antinuclear, and antiphospholipid antibodies were negative. The pathergy test was positive.

During his stay at the hospital, the patient developed genital and oral ulcers (figure 2) consistent with BD.

Figure 2. Oral (a) and genital (b) aphthosis

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And he also developed right superficiel femoral thrombosis confirmed byDoppler ultrasonography and CT-angiography (figure 3). At this stage, our patient was diagnosed with BD according to the International Criteria for Behcet's disease with a score of 6.
The patient received corticosteroid, colchicine and immunosuppressive therapy, with good clinical and biological progress.

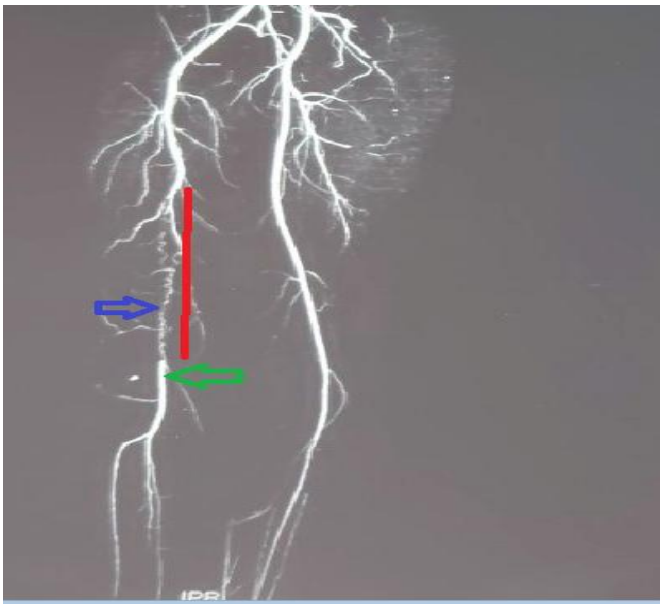

Figure 3. Totally thrombosed right superficial femoral artery (red line). The popliteal and tibial arteries are opacified and of good caliber (green arrow) thanks to well-developed collateral circulation (bleu arrow).

Discussion

Behcet's disease, a multisystem disease, is classified among vasculitides by the 2012 Revised International Chapel Hill Consensus Conference Nomenclature of Vasculitides, in the group of Variable Vessel Vasculitis However, some authors, still classify the disease among the auto-inflammatory diseases (3). The disease is mostly encountered at the third and fourth decades of life (4). The highest prevalence is found in Turkey, with up to 420 per 100,000 persons affected. Iran, northern China, and Korea follow with the next highest prevalence (1, 5). The diagnosis of Behcet's Disease, in the absence of a characteristic biological test, is clinical and upon an expert opinion. However, Classification/Diagnosis (C/D) criteria may be of help. It is interesting to note that no disease has ever had so many Classification/Diagnosis criteria as BD (3,5). These criteria require the presence of oral ulcers, in addition to two or more of the following manifestations: genital ulceration, eye lesions (uveitis, retinitis), skin lesions (folliculitis, papulopustular lesions, acneiform nodules, erythema nodosum), and a positive pathergy test (6). In addition, patients with Behçet's disease can present with other symptoms, such as thrombophlebitis, deep venous thrombosis, central nervous system involvement, arthralgia, arthritis, and gastrointestinal features (5). Vascular involvement is one of the most important features of BD, although not clinically seen in all patients (3); vascular manifestations were seen in 9.1% of patients (9) and has been reported to occur in 14.7% - 27.7% of BD patients (3, 7). Vasculo-BD was male-predominant (62%), whereas females predominated in the other BD subtypes (female 56%) (8).
The arterial involvement is seen in 3 to 5% (5); it has drawn special attention due to its significant association with mortality, as this complication accounts for over a quarter of all BD-related deaths (2). Arterial thrombosis was seen 0.154% of cases and aneurysms 0.5% of cases (3, 7); aneurysms were the first presentation in 26% of the patients with arterial aneurysm, while the remaining 74% developed after the onset of disease by 3.2 ± 3 years (9). The abdominal aorta is the most common site for rapidly growing aneurysms accounted for approximately 60% of all arterial involvement in Behcet (8). Abdominal pain is the leading symptom. An arterial aneurysm is a focal dilation of a blood vessel with respect to the original artery. Vasculitis of the vasa vasorum of large arteries and necrosis of the vascular wall occur, which are followed by

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spurious aneurysms (4). Aneurysms in Behcet's disease are thought to result from obliterative endarteritis coupled with intense inflammatory process resulting in destruction of the media and fibrous thickening of both intima and adventitia thereby distending the arterial wall. The pathogenesis of aneurysms in Behcet's syndrome is due to the destructive inflammatory process. Matrix metalloproteinase proteins and especially MMP-9 may play an important role as was reported on (8). Treatment options are unspecific and aim to relieve symptoms and control disease progression and severity. Disease management usually includes systemic anti-inflammatory and/or immune modulating drugs (1-2, 5, 10). Conventional open surgery is the most commonly used treatment for the arterial lesions in Behçet's disease patients. The vascular anastomosis can be wrapped and reinforced by using prosthetic materials such as Polytetrafluorethylene or Dacron ®as a patch graft (4, 10).

Conclusion

Behçet's disease is a systemic vasculitis of unknown etiology, characterized by recurrent oral and genital ulcers and uveitis. Although the etiology and pathogenesis is not clearly defined, genetic predisposition, infections and immunological dysfunctions have been implicated. Occlusion and aneurysms of major arteries commonly lead to bleeding, infarction and organ failure, particularly in instances of pulmonary aneurysm. Rupture of aneurysms may be fatal.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

Authors' contributions

The authors contributed to the content, conception, and design. Nassima Dekdouk was the major contributor to writing the manuscript.

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