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Research Article

Peripheral Ischemia and Necrosis Revealing Buerger's Disease in an Internal Medicine Department

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Abstract

Buerger's disease is a systemic vasculitis strongly correlated with tobacco use. It entails a heavy socio-occupational impact. We report the case of a 26-year-old patient with ischemia of the lower limbs revealing Buerger's disease. He is known smoker with passive exposure to Indian hemp. He was seen for ischemic foot pain that had been progressing for a year. Involvement began in the left big toe. Subsequently, hyperalgesic necrotic lesions were observed on the 3rd, 4th and 5th toes of the foot. Biological investigations revealed an inflammatory syndrome with normocytic anemia and increased C-reactive protein. Retroviral, syphilitic, Hepatitis B and C viruses and SARS-CoV-2 serologies were negative. Antinuclear antibodies were initially borderline at 100 IU with speckled fluorescence, then negative on control. Neutrophil cytoplasmic antibodies and antiphospholipid antibodies were negative. Investigation of thrombophilia was non-contributory, notably factor V mutation testing, antithrombin III assay, proteins C and S and fibrinogen. An arterial ultrasound revealed extensive arterial thrombosis with thickening of the femoral arterial vessel walls. Thromboangiitis obliterans was confirmed and the patient was put on corticosteroids and adjuvant therapy. Surgical treatment was performed 4 months later. Buerger's disease is a serious vascular disorder which must be diagnosed very early in order to prevent complications. Early smoking cessation leads to remission in the early phase.

Keywords

Thromboangiitis Obliterans, Buerger's, Tobacco, Amputation

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1. Introduction

Buerger's disease or thromboangiitis obliterans is a systemic vasculitis characterized by inflammatory, non-atherosclerotic vascular occlusive disease with thrombosis and recanalization, involving small and medium-caliber veins and arteries of the upper and lower extremities [1]. It is a rare condition, with a prevalence of 1/10000 in Europe; it is present in other parts of the world, but seldom described. It is closely linked to active or passive smoking [2]. We report the case of a 26-year-old man with no specific pathological history who was treated for occlusive disease of the lower limbs with distal necrosis revealing thromboangiitis obliterans.

2. Observation

The patient was 26 years old, a known smoker at 11 pack-years, with a history of passive intoxication with Indian hemp, non-alcoholic, non-diabetic and without any other cardiovascular risk factors. For the past year, he had been experiencing intense pain exacerbated by prolonged walking, even at rest. This painful symptomatology progressed by flare-ups and remissions. Involvement began in the left big toe (Figure 1). Subsequently, hyperalgesic necrotic lesions were observed on the third, fourth and fifth toes of the right foot. He consulted several times in surgery and general medicine. He refused any radical treatment. Initial surgical treatment was offered in a health facility, with some stabilization of the lesions. Three months later, a relapse was observed, and on clinical examination the patient presented with a necrotic wound over the entire left foot, extending to the ankle (Figures 2 and 3), associated with abolition of distal pulses and cooling of the limb. An arterial Doppler ultrasound revealed extensive arterial thrombosis with thickening of the femoral arterial vessel walls.

Biological investigations revealed an inflammatory syndrome with normocytic anemia and increased C-reactive protein. Retroviral, syphilitic, B and C viral and SARS-CoV-2 serologies were negative. Antinuclear antibodies were initially borderline at 100 IU with speckled fluorescence, then negative upon control. Neutrophil cytoplasmic antibodies were negative. Antiphospholipid antibodies were negative. Investigation of thrombophilia was non-contributory, notably factor V mutation testing, antithrombin III assay, proteins C and S and fibrinogen.

Following these investigations, thromboangiitis obliterans was diagnosed. The patient was started on prednisone 1mg/kg/day with adjuvants and acetylsalicylic acid.
An amputation was initially proposed to the patient but he was not convinced. As the necrotic lesions progressed, surgery was performed and the necrotic tissue was removed. These days, we are observing a good evolution of the symptomatology with no new lesions on the limbs. The patient showed stabilization of the lesions with no recurrence of other signs.

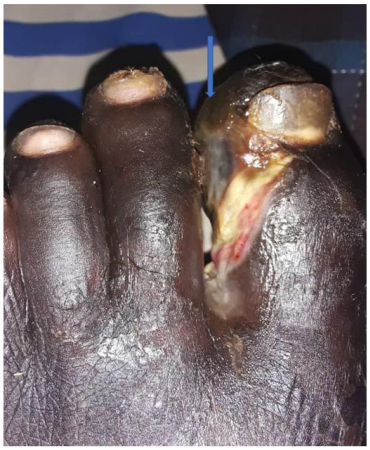

Figure 1. *Ischemia and necrosis of the left big toe.*

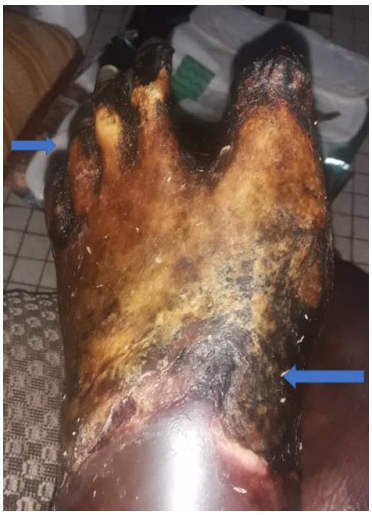

Figure 2. *Necrotic lesion of the left foot in thromboangiitis obliterans.*

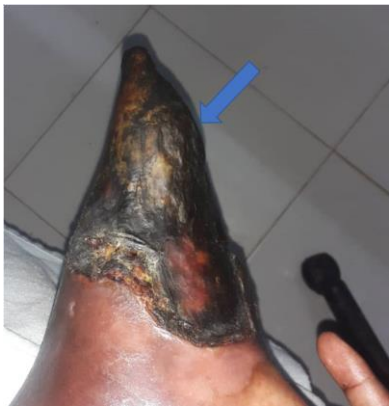

Figure 3. *Profile view of left foot necrosis.*

3. Discussion

Buerger's disease is a rarely described vasculitis in sub-Saharan Africa. It results in non-atherosclerotic inflammatory vascular obstruction. It is a very rare condition. It mainly affects men under 40 who smoke or are passively exposed to tobacco. It involves small and medium-calibre vessels in the legs and arms [3]. It was most frequently described in the Middle East and India, followed by Europe. Its prevalence among all vasculitides ranged from 0.5 to 5.6% in Western Europe [4]. There are few descriptions of this disease in Africa. Initial symptoms include numbness, heaviness and sometimes paresthesia, before the onset of ischemic signs. The latter can lead to necrosis if not diagnosed early. This leads to all the psychological complications observed in these patients. A long period of denial follows, which may lead to the delay in treatment observed in our patient [5].

This is a pathological entity that is well differentiated within the scientific community. The link with smoking is well documented. Cessation of smoking in the early stages of the disease would reduce disease progression and avoid limb amputations in patients. The pathophysiology of the disease involves three phases: acute, sub-acute and chronic. It associates an infiltrate of polymorphic leukocytes, predominantly neutrophils, creating microabscesses in thrombi, sometimes with granulomatous inflammation, sometimes followed by a phase of fibrosis [4]. Medical imaging is of major help in assessing the degree of stenosis. Doppler ultrasound, CT angiography and sometimes capillaroscopy can be used for small digital vessels [6].

Diagnosis is based on a number of criteria. There are several, but the Shionoya criteria are the most widely used. These involve a history of smoking, whether active or passive, age under 50, occlusion of the sub-popliteal arteries, involvement of the upper limbs and notion of migratory phlebitis, with exclusion of all other atherosclerosis risk factors except smoking. However, it is advisable to rule out other vasculitis etiologies before adopting the diagnosis. These include inflammatory, lipid and hemostasis assessments, as well as immunological tests (cryoglobulinemia, anti-phospholipid, ANCA, antinuclear antibodies) and viral serologies (HIV, HVC, HVB) [7].

In tropical Africa, this condition can be confused with Hansen's disease. The latter is considered a neglected tropical disease. It is a serious, mutilating condition caused by mycobacterium laprae [8]. The other difficulty lies in our knowledge of other systemic vasculitis, which are not described enough in Sub Saharan Africa. A series of 27 cases was published by Ndongo et al in 2010. These affections are underdiagnosed due to lack of adequate resources in developing countries [9].

To date, no single etiology has been identified. However, several theories have been put forward, including the presence of immunological factors with impaired endothelium-dependent vasodilation. It differs from most vasculitides in that the thrombus is very rich in inflammatory cells, with relative respect for the vascular walls. Markers of acute inflammation are often normal. There is almost no evidence of immune activation such as complement, cryoglobulinemia, antinuclear antibodies, rheumatoid factor... It is a diagnosis of exclusion [10].

There are atypical manifestations of Buerger's disease. Consequently, a good knowledge of this condition is essential. Lower limb involvement is not exclusive, as other organs or viscera may also be involved. Digestive manifestations such as abdominal pain and bloody diarrhea without biological inflammatory syndrome revealing ulcerative colitis have been reported in a young subject [11].

These digestive disorders are what make this condition so serious. Another case of mesenteric infarction in a 37-year-old has even been reported [7].

Treatment must be initiated as early as possible, before ischemic complications arise. It is difficult to diagnose this condition at this stage, which means that professionals need to be well-trained in the disease. In most cases, a ban on smoking halts the progression of symptoms and preserves ischemic limbs. Corticosteroid therapy helps achieve remission. In severe cases, such as necrosis with risk of extension, amputation is recommended [3, 4, 8].

4. Conclusion

Buerger's disease is a serious vascular disorder which must be diagnosed very early in order to prevent complications. Diagnosis requires elimination of other causes of vascular occlusion. There are few descriptions from sub-Saharan Africa. Internal medicine specialists should be more vigilant in the initial diagnosis of this form of vasculitis. Early smoking cessation leads to remission in the early phase.

Abbreviations

CT	Computed Tomography
ANCA	Antineutrophil Cytoplasmic Antibodies
SARS-CoV-2	Severe Acute Respiratory Syndrome Coronavirus 2
HIV	Human Immunodeficiency Virus
HCV	Hepatitis C Virus
VHB	Hepatitis B Virus

Author Contributions

Diallo Bachir Mansour: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Resources, Writing – original draft, Writing – review & editing

Jean Noel Ndour: Formal Analysis, Methodology

Ndiaye Yanidou: Formal Analysis

Faye Fulgence Abdou Faye: Formal Analysis

Moustapha Mbacke Diop: Investigation, Resources
Aboubakry Sow: Formal Analysis
Nana Mamoudou Baba: Formal Analysis, Writing – review & editing
Wade Adama: Formal Analysis
Berthé Adama: Data curation, Supervision
Touré Papa Souleymane: Data curation, Methodology
Diop Madoky Magatte: Data curation, Methodology, Supervision
Ka Mamadou Mourtalla: Supervision, Validation

Conflicts of Interest

The authors declare no conflicts of interest.

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