



Case Report

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Recurrent and resistant duodenal ulcer revealing Thromboangiitis obliterans

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Abstract

Thromboangiitis obliterans or Leo Burger's disease is a rare systemic vasculitis in young and typically heavy smoker male, whose diagnosis is a real challenge for the clinician. Digestive manifestations are rare and unusual in this disease and the inaugural intestinal involvement is extremely found. We report an original case of relapsing and resistant duodenal ulcer revealing a Leo Buerger's disease in a 42 year old man.

Keywords: Thromboangiitis obliterans, Leo Buerger's disease, Duodenal ulcer, Vasculitis.

INTRODUCTION

Described first in 1905 by Leo Buerger^[1], Thromboangiitis obliterans is a rare systemic vasculitis in young and typically heavy smoker male. Its diagnosis is a real challenge for the clinician. The anatomical substrate is a segmental and multifocal inflammation of arterial, veins and nerve structures resulting in a real panvasculitis^[1].

Digestive manifestations are rare and unusual in this disease^[2] and the inaugural intestinal damage is exceptionally found^[3].

We report an original case of relapsed and resistant duodenal ulcer revealing a Leo-Buerger's disease.

CASE REPORT

42-years-old patient, heavy smoker (40 cigarettes per day for more than fifteen years) who presented a recurrent duodenal ulcer lasting for five years and not improved by the well-behaved anti-ulcer treatment. This duodenal ulcer was confirmed by endoscopy three times for three successive years. Histological examination, performed twice, was non-specific (deep ulcerations associated with microthrombosis and absence of *Helicobacter pylori*). It was not noted a suspicious drug intake or non-adherenceto prescribed anti-ulcer treatment. The search for an underlying solid cancer or hematologic malignancy was negative. Also, it has not been noted any clinical or laboratory evidence for underlying systemic vasculitis or autoimmune disease.

One year after the last episode of duodenal ulcer, he was hospitalized for evaluation of recurrent superficial phlebitis of the right leg (twice a month) associated with recent intermittent claudication of both lower limbs. The distal pulses were absent in the left lower limb. The vascular morphological imaging (ultrasound coupled with Doppler, arteriography and venography) showed the association of phlebitis and arteritis: thrombosis of the right femoral vein, thrombosis of the right great saphenous vein, occlusion of the left anterior tibial artery, the right posterior tibial artery and the left dorsalis pedis artery associated with stenosis of the left posterior tibial artery and the right dorsalis pedis artery. The biological investigations were within the normal range. The immunological tests did not reveal any significant abnormality (anti-nuclear antibodies, anti-native-DNA antibodies, anti-soluble antigens antibodies, anti-neutrophil cytoplasmic antibodies (both cytoplasmic and perinuclear variants) and anti-phospholipids antibodies). The search for a hereditary or acquired thrombophilia was negative (prothrombin time, activated partial thromboplastin time, anti-thrombin III deficiency, protein C deficiency, protein S

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Sdeficiency, and the Leiden variant of factor V).

Thus the diagnosis of Thromboangiitis obliterans was retained. The evolution was quickly marked by the installation of a dry distal gangrene leading to iterative amputation of the fifth and the first toes of the left foot. With effective anticoagulation, peripheral vasodilators and tobacco cessation, the subsequent evolution was satisfactory.

During his hospitalization and because of persistent epigastric pain, upper gastrointestinal endoscopy was performed objectifying rounded duodenal ulcer of 5 mm, from the anterior surface of the first duodenum associated with congestive gastritis and superficial ulcerations of the pre-pyloric antrum. Histological examination showed a deep and digging duodenal ulcer associated with multiple micro-thrombosis and polymorphic inflammatory infiltrate (predominantly lymphocytic). With classic anti-ulcer treatment, in addition to the vasodilator and anticoagulant therapy of Buerger's disease, evolution was made to the complete healing of digestive ulcer confirmed by endoscopy control made two months later. No ulcer recurrence was observed for four years.

DISCUSSION

Thromboangiitis obliterans is a rare distal angiopathy representing less than 4-5% of all ischemic vasculopathies^[4]. It has long been controversial, and it is only in 1962 that it was recognized as a distinct clinic-pathological entity and was eventually separated from arteriosclerosis obliterans.

It is now known to be a systemic vasculitis predominant in young and heavy smoker males. It preferentially affects neurovascular structures (arteries of medium and small size, veins and nerves) leading to true panvasculitis with segmental and multifocal involvement^[1] and making its diagnosis a challenge for physicians^[2,3].

The association between Leo Buerger's disease and peptic ulcer was noticed long ago^[5,6], but it remained too controversial^[5]. These ulcers can affect the entire digestive tract: stomach, duodenum^[6], jejunum^[3], ileum^[2] and colon^[7]. They are often multiple^[2], rarely unique^[3], and are frequently complicated by perforation^[2,3].

The vascular involvement has been suggested as a pathophysiological mechanism of peptic ulcers in thromboembolic obliterans, but it is only recently that the ischemic nature of these ulcers has been proven. These ulcers are related to digestive micro-thrombosis proven radiographically and histologically^[3].

These intestinal complications of Thromboangiitis obliterans can be acute or chronic, remain asymptomatic^[8], or exceptionally be the first symptom revealing the disease^[3]. Similarly, a correlation between the presence of peptic ulcer and the severity of the disease is also reported^[9].

This association raises once again the pathogenic role of tobacco in the genesis of these two conditions; in fact, the involvement of smoking in the peptic ulcer disease is now proven in large population studies^[10].

CONCLUSION

Peptic ulcer is an exceptional mode of revelation of Leo-Buerger's disease. This association deserves to be known and justify the search of Thromboangiitis obliterans in any patient with relapsed and/or resistant gastro-duodenal ulcer, especially when occurring in young and heavy smokers males.

Conflicts of Interest: No conflicts.

Authors contributions

Drafting the article: Salem Bouomrani, revising it: Maher Beji, all authors had participated in the management of this case. All authors read and approved the final version of the manuscript.

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