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Case Report

Buerger's disease in female: rare case report

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ABSTRACT

Thromboangiitis obliterans (Bureger's disease) is a vascular disease characterized by a segmental, non-atherosclerotic inflammation of the small and medium-sized arteries and veins of the distal extremities mostly affecting young male smokers and rarely females. Only a few cases with histologic proof of the disease in females have ever been recorded in literature. We report a rare case of Buerger's disease in a 55 years old female.

Keywords: Inflammation, Non-atherosclerotic, Thromboangiitis obliterans

INTRODUCTION

Thromboangiitis obliterans (TAO) is a clinical syndrome characterized as a miscellaneous form of nonatherosclerotic vasculitis affecting small and medium-sized arteries and veins. TAO causes painful ischemic ulcers of the digits and gangrene of the toes and fingers as a result of the vascular ischaemia. Afflicted patients are mostly young, male, inveterate tobacco smokers.[1] Although the exact underlying causes of Buerger's disease are still unknown, the disease is strongly associated with tobacco smoking.[2] The disease is more prevalent in India, Southeast Asia and eastern European countries than the West.[3]

CASE REPORT

A 55 years old female, nonalcoholic, non-diabetic; presented with severe rest pain in the both lower limb since last 6 months and blackish discoloration of right lateral two toes for 1 week. The patient is a chronic smoker for past 20-years, smoking 10 cigarettes a day. She also gave a history of gradually worsening claudication pain in both lower limbs, more on right side since last 10 years.

General examination

Pulse 75/minutes, regular, normovolumic, BP 110/70 mm Hg, blood pressure in both arms are symmetrical, JVP not raised, RR 20 breaths/ minutes.

Figure 1: Dry gangrene of right 4th and 5th toes with a non-healing ulcer on the heel.

Local examination revealed dry gangrene of 4th and 5th toes of right foot and non-healing ulcer on the heel with signs of ischemia in the form of dryness of the skin, loss of subcutaneous fat, loss of hair and brittle nails as shown in Figure 1. Examination of opposite leg showed absence of 4th toe

Examination of peripheral pulsations of lower limbs showed absence of infra-popliteal pulsations as shown in Table 1. All peripheral pulsations of upper limb were present.

Table 1: Examination of peripheral pulsations of lower limbs.

Pulse	Femoral A	Popliteal A	Anterior tibial A	Posterior tibial A	Dorsalis Pedis A
Right leg	Present	Absent	Absent	Absent	Absent
Left leg	Present	Absent	Absent	Absent	Absent

Table 2: Blood investigations result.

Investigations	Result
Hemoglobin	9.4 gm/dl
ESR	35 mm/hr
Total leucocyte count	8.8 × 109/L
Platelet count	2,30,000/dl
Differential count	
Neutrophils	70%
Lymphocytes	26%
Monocytes	2%
Eosinophils	2%
Bleeding time	1'30"
Clotting time	4'20"
Hematocrit	37%
CRP	Negative
VDRL	Non-reactive
RBS	110 mg/dl
Serum Bilirubin	0.2 mg/dl
Serum Creatinine	0.7 mg/dl
Fasting plasma lipid profile	
Total CHOLESTEROL	208 mg/dl
Triglycerides	310 mg/dl
LDL	117 mg/dl
HDL	27 mg/dl
ELISA for ANA	Negative
Serum Homocysteine	5.73 micromol/L

Special tests

Pain and pallor appeared on raising of leg in Buergers test. Buerger's angle was found to be 40-degree on right side and 50-degree on the left. Sensory and motor systems of both lower limbs were normal.

Doppler study report of lower limb arterial system

Both right and left limb showed similar pattern. Tardus Parvus wave pattern seen in common femoral, superficial and deep profunda femoris, popliteal, anterior tibialis artery; dorsalis pedis shows no arterial flow pattern as shown in Figure 2. Common femoral artery showed irregular wall thickening (intima media thickness 2.8 mm) with echogenic calcified lesion measuring 4.3 mm.

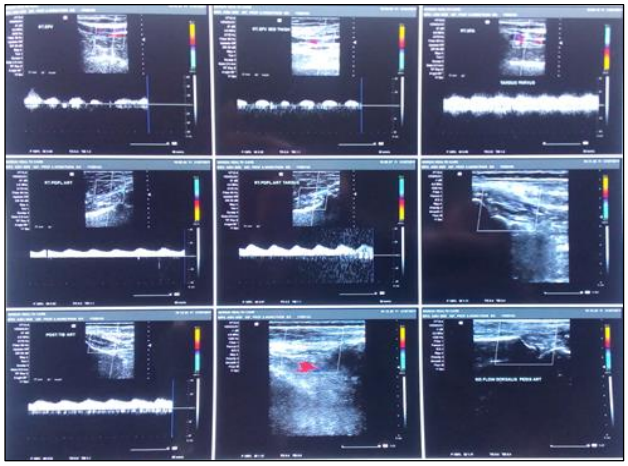

Figure 2: Doppler study of lower limbs arterial system.

Histopathological report

Specimens were taken for histologic examination from the disarticulated specimen of gangrenous right 4th and 5th toes and surrounding skin. Medium and small size arteries displayed thrombosis (Figure 3) without any evidence of recanalization.

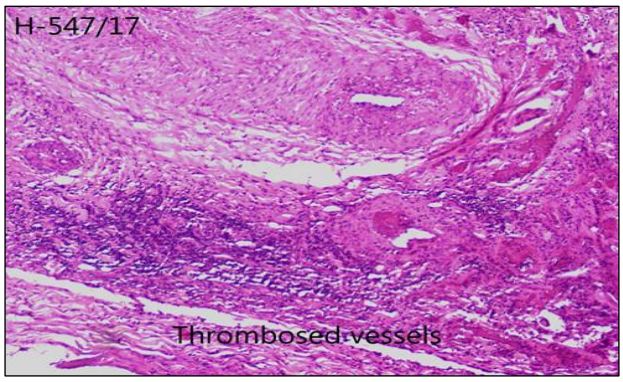

Figure 3: Thrombosed vessels.

Moderate amount of mixed perivascular inflammatory cell infiltrate predominantly polymorphonuclear cells, suggesting acute phase lesion of Thromboangiitis Obliterans (Figure 4).

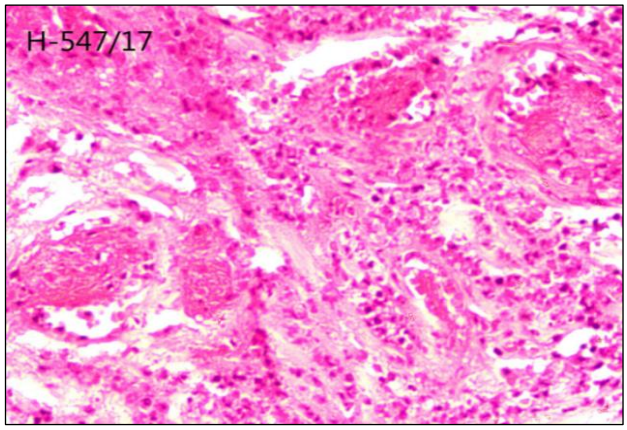

Figure 4: Perivascular inflammatory cell infiltrates.

Management

Initially patient was managed conservatively with oral aspirin for pain relief, antibiotics according to culture and sensitivity and daily dressing of right heel ulcer. Total abstinence from smoking was advised. Disarticulation of gangrenous right 4th and 5th toes was performed.

Outcome and follow-up

Patient was followed up every month for six months. The combination therapy with anticoagulation strategy, aspirin, prednisolone, pentoxifylline, nifedipine and atorvastatin were prescribed. Adherence to smoking withdrawal was advised.

DISCUSSION

The traditional diagnosis of Buerger's disease is based on 5 criteria (smoking history, onset before the age of 50 years, infrapopliteal arterial occlusive disease, either upper limb involvement or phlebitis migrans, and absence of atherosclerotic risk factors other than smoking). As there is no specific diagnostic test and an absence of positive serologic markers, confident clinical diagnosis should be made only when all these 5 criteria have been fulfilled although not universally accepted. In our patient also these 5 criteria were not fulfilled but clinical presentation was consistent with Buerger's disease with histological proof. Exclusion of other diseases, e.g. atherosclerotic occlusive disease, collagen disease, and hypercoagulopathy, is necessary. Buerger's disease is becoming more common in women.[4,5] Njo KT and Smit AJ reported Thromboangiitis Obliterans in two female smokers.[6] Olin J et al in his study reported 23% female patients of TAO.[7] Yorukoglu Y et al also reported an increased incidence of TAO in women.[8] In recent reports, 11% to 23% of patients were women it may be due to increased use of tobacco by women.[9] The clinical presentation and histopathology of TAO in women differ in no way from that in men. There is no evidence that the disease is milder in women.

CONCLUSION

Awareness of the entity and familiarity with the clinical, angiographic, and pathologic features of the disease are the key to a prompt and correct diagnosis of Buerger's disease. The diagnosis of Buerger's disease may be missed in patients with old age or female gender and should not be excluded because of these patient characteristics. Currently, there is no specific treatment for TAO. Absolute discontinuation of tobacco use is the only strategy proven to prevent the progression of Buerger's disease.

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