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Intestinal ischaemia in Buerger's disease in a patient presenting with lower limb gangrene: Case Report and Literature Review

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INTRODUCTION

Buerger's disease was described and established in the English literature in 1908 as an entity distinct from atherosclerosis1. The disease usually affects medium and small arteries and veins of the upper and lower extremities. Visceral artery involvement is rarely reported. It is a disease of the orient. Its incidence varies from 10-33% in Japan and Bombay. Similarly high incidence rates have been recorded in South East Asia2,3,4. We report a case of Buerger's disease with intestinal ischaemia caused by SMA occlusion.

CASE REPORT

A 50 year old chronic smoker presented to emergency department with severe abdominal pain with muscle guarding. Pain was out of proportion to clinical signs. Emergency laparotomy was performed for suspected ischaemic intestinal involvement. At laparotomy, whole of the ileum and caecum and proximal side of ascending colon were gangrenous. Gangrenous small and large intestines were resected and end jejunostomy was created.

Post operative nutrition was maintained with total parenteral nutrition. Patient was orally started after 11 days. Once, haemodynamics was established patient was discharged on 20th post op day.

Patient returned to us after 1 month with gradually progressive painful discolouration of right lower limb. Lower limb pulses were not palpable except for the femoral artery. Pulses in the upper extremities were palpable. The patient had smoked about 40 cigarettes per day for 20 years, but had no other risk factors for atherosclerotic disease, for example:- hypertension, hyperlipidaemia, diabetes mellitus.

CT angiography showed complete transmural segmental thrombotic involvement of popliteal, anterior tibial, posterior tibial and dorsalis pedis vessels on right side. Left lower limb arteries showed partial occlusion. There was no collateralization of lower limb arteries of lower limb vessels. Patient stopped smoking only during hospitalization and he was started with daily oral anticoagulant medicine, pentoxiphylline and aspirin. Lower limb gangrene worsened, subsequently below knee amputation was performed.

Histolopathological findings revealed that the posterior tibial artery was affected, compatible with Buerger's disease. Pathologic examination revealed that the posterior tibial artery had intact elastic lamina, with organized thrombi, and three layers preserved almost intact.

DISCUSSION

In 1908, Buerger's described first case of thromboangitis obliterans (Buerger's disease) and reported that this obliterating vasculopathy could involve visceral arteries. However this uncommon vasculopathy mainly affected the medium and small arteries in the extremities, and visceral cerebral arteries were rarely affected.

Our patient had rare intestinal ischaemia caused by Buerger's disease. The patient had typical symptoms of Buerger's disease at the second visit. He had no atherosclerotic risk factors except for tobacco abuse, and no serologic data suggested any collagen disease or anticoagulation disorder. An electrocardiogram revealed no atrial fibrillation that could cause distal embolization. An angiogram of lower limb showed findings typical of Buerger's disease, e.g. abrupt occlusion and tree root pattern. No typical pattern of Buerger's disease was found on abdominal aortogram. However, pathologic examination demonstrated that the arteries of the resected small intestine had organized thrombi with an essentially intact elastic lamina and inflammatory cell infiltration. Intestinal Buerger's disease demonstrates the same histologic findings as in arterial and venous lesions. Whereas intestinal Buerger's disease is rare, ischaemia of upper and lower extremities is the most common clinical manifestation.

Clinical criteria for diagnosis of Buerger's disease are smoking, onset of symptoms before the age of 50 years, infrapopliteal arterial occlusive lesions, either upper limb involvement of phlebitis migrans, absence of atherosclerotic risk factors except smoking.

Besides the extremities, Buerger's disease has also been reported to sometimes affect cerebral, coronary, internal thoracic, renal and mesenteric arteries4. Mesenteric vascular occlusion is a life threatening cause of bowel ischaemia which needs prompt and accurate diagnosis. Mesenteric vessel involvement is rare and the exact incidence of this manifestation has not been established. In most cases it develops after the onset of peripheral vascular disease, whereas mesenteric ischaemia as an initial presentation of Buergers is extremely rare, with only 5 such cases published in the English literature. Three of these patients continued to smoke and subsequently developed peripheral vascular disease; one patient stopped smoking and did not develop peripheral vascular disease and one patient died postoperatively. The clinical presentation of mesenteric ischaemia in Buergers disease is not specific and encompasses acute or chronic abdominal pain, diarrhea, weight loss, intestinal occlusion or acute peritonitis requiring prompt surgical intervention. The nonspecific presentation poses a diagnostic challenge. Even after identifying mesenteric ischaemia as the cause of gastrointestinal symptoms, establishing the correct diagnosis requires a thorough clinical, radiological, laboratory and histopathological work up5.
The differential diagnosis of mesenteric ischaemia, ordered according to the principal underlying the mechanism is shown in table 1.

Table 1: Causes of mesenteric ischaemia according to pathogenesis

1. Thrombo-embolic causes	Cardiac thrombus. Cholesterol emboli. Atrial myxoma. Anti-phospholipid syndrome. Hyper-coaguable states(e.g.- protein C and S deficiency) Atherosclerosis
2. Vasculitis	
Primary type	Polyarteritis nodosa. Wegners granulomatosis Microscopic polyangitis. Cryoglobulinemia. Buergers disease. Churg strauss syndrome.
Secondary type	SLE. Rheumatoid arthritis. Myelodysplastic syndromes. Haematological malignancies. Infections
3. Mechanical obstruction	Strangulated hernia. Volvulus. Aortic dissection. Intussusception. Tumour compression. Superior mesenteric artery syndrome. Median arcuate ligament syndrome.

According to the recent data by Eichhorn et al anti-endothelial antibodies might be useful for the diagnosis of Buerger's disease. In this study, serum anti-endothelial cell antibody titres were significantly higher during active Buergers disease compared to patients in remission ($p<0.01$), and also significantly higher in patients during remission compared to healthy subjects ($p<0.001$). Additional studies are required to further delineate the sensitivity and specificity of this test.

Smoking tobacco is essential for the initiation and progression of Buergers disease, and most patients diagnosed with Buergers disease are heavy smokers. Indeed the only proven strategy to prevent progression of disease and avoid amputation is the complete discontinuation of tobacco smoking or other use of tobacco in any form6.Pedical omental transfer was proposed as a mode of treating these patients, particularly when limb ischaemia is present and excellent results have been reported. Traditionally, it has been performed only after failure of lumbar sympathectomy7. The property of omental pedicle to induce neo-angiogenesis and thus improve circulation of surroundings tissues has been well established. A lipid angiogenic factor from omentum has been proposed to be responsible for this property of omentum8. Surgical revascularization, spinal cord stimulation omental transfer, immunoglobulin A immunoadsorption and intramuscularly administered vascular endothelial growth factor (VEGF) gene therapy are other options that have been used to aid ulcer healing and pain relief9.

TAKE AWAY MESSAGE

Mesenteric ischaemia is a serious complication of Buerger's disease that occur at any stage of the disease. Chronic mesenteric ischaemia as an initial manifestation of Buerger's disease is very rare and is often diagnosed very late ,i.e. after serious complications such as malabsorption and/or acute ischaemia and necrosis have already occurred. A high index of suspicion for this disease is needed to enable early diagnosis of mesenteric ischaemia in a heavy smoker. During the course of this disease intestinal involvement is possible, and careful observation for abdominal manifestations is mandatory.

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FIGURES USED

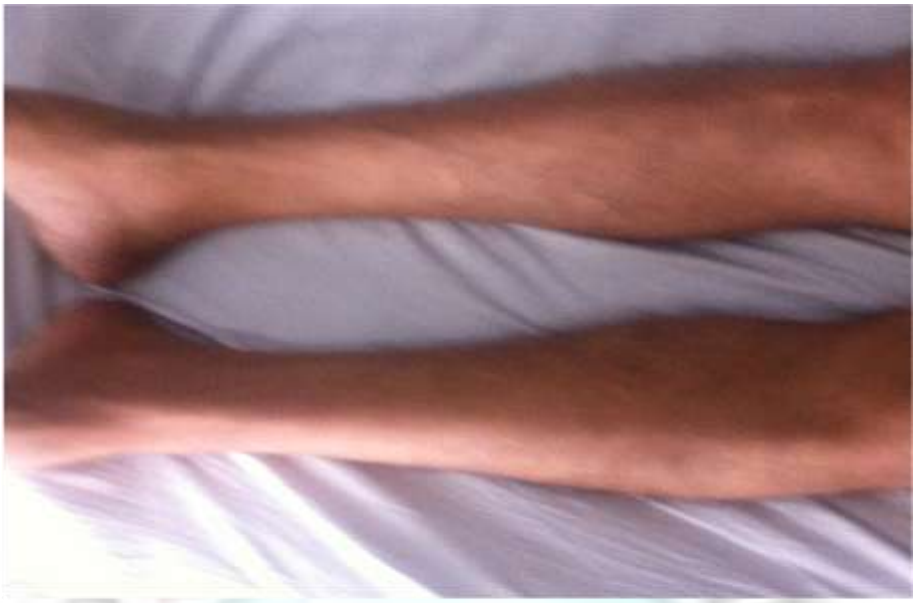

Fig 1. Buerger's Disease of lower limb

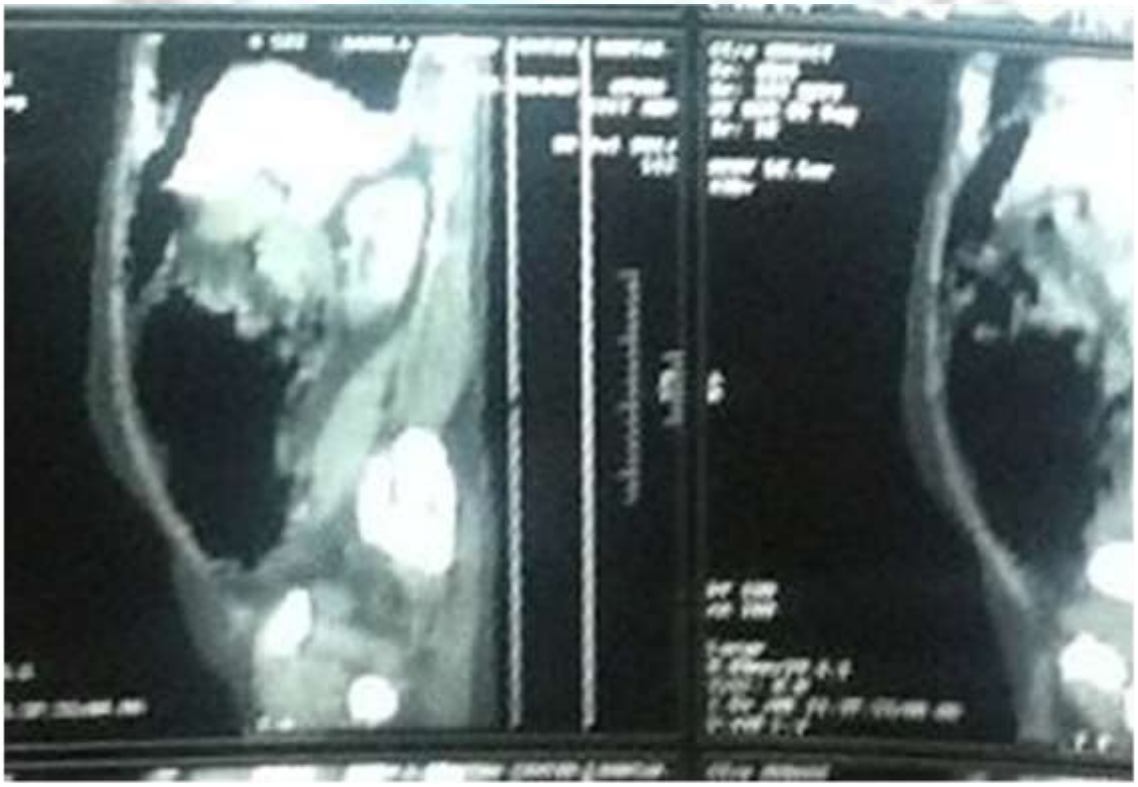

Fig 2. CT Angiography of Superior Mesenteric Art.

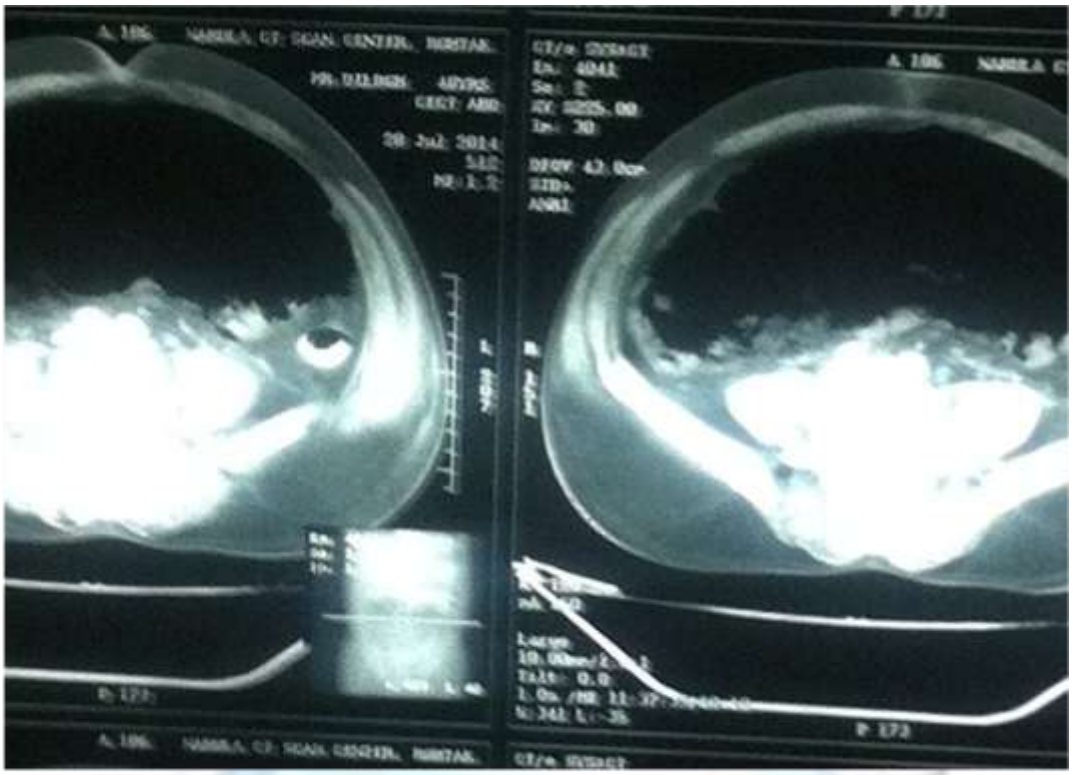

Fig 3. CT abdomen of intestinal Ischemia

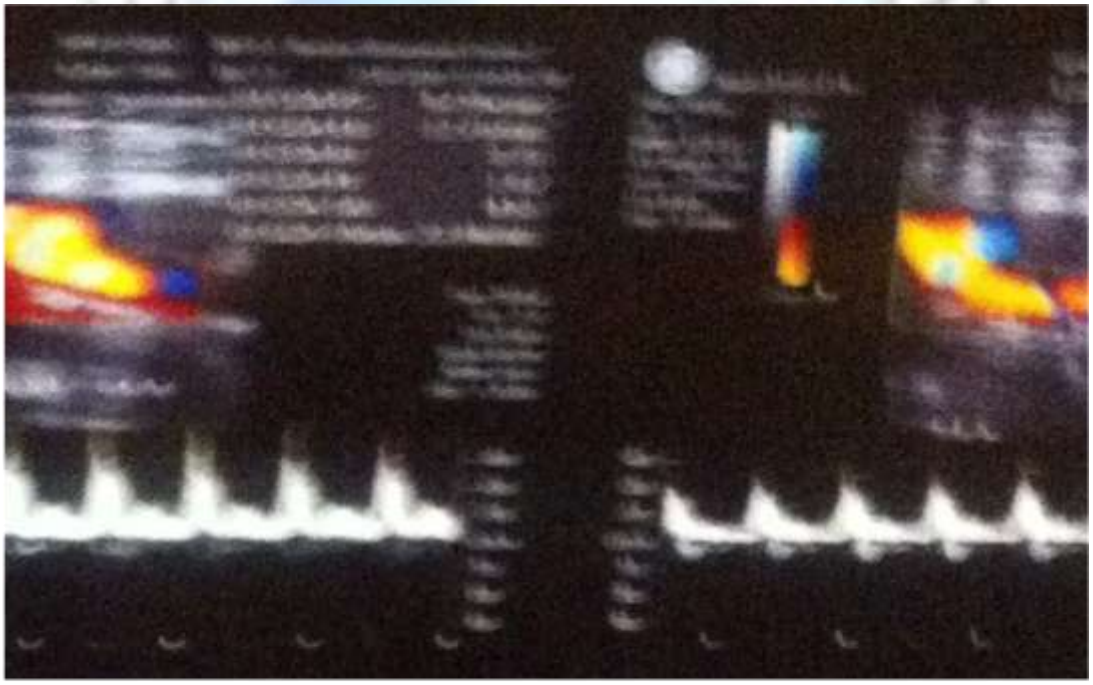

Fig 4. Color Doppler of left lower limb

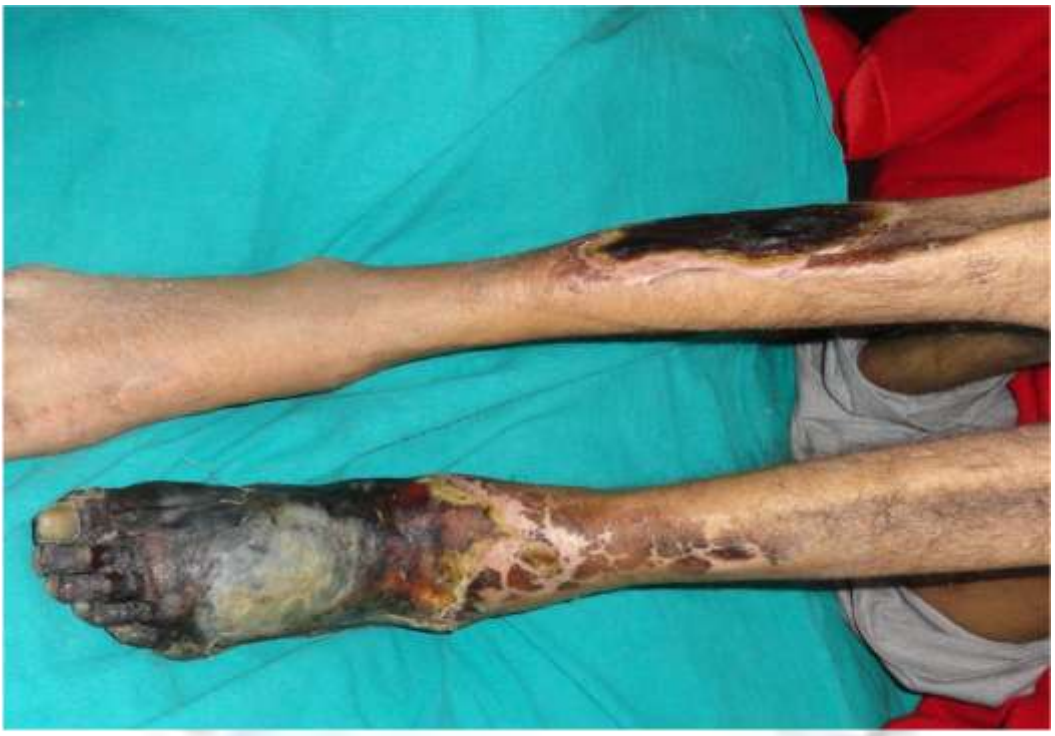

Fig 5. Lower limb gangrene