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Case Report

Cutaneous Manifestations of Aortoiliac Occlusive Disease: Two Cases and Review of the Literature

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Abstract

Aortoiliac occlusive disease (AIOD) is non-inflammatory obstructive vasculopathy commonly affecting patients with advanced atherosclerosis, diabetes mellitus, or elevated cholesterol levels, and subjects with other risk factors such as cigarette smoking.

Two Caucasian patients (a 55-year-old woman and a 56-year-old man), with ulcerous cutaneous lesions of AIOD are reported. In both cases, medical history comprises initial lower limb claudication, multiple painful ulcers along the legs and absence of superficial femoral artery pulse. Severe obstruction of both infrarenal aorta and iliac arteries on the left side was demonstrated by contrast angiography and Doppler ultrasonography.

The evolution of the disease showed some characteristic findings, including pyoderma gangrenosum-like ulcerations as the initial cutaneous manifestation of AIOD, multiple painful ulcers along the lower extremities, and aorto-iliac occlusive disease due to atherosclerosis.

Early diagnosis and surgical reconstruction of vessels in patients with AIOD improved quality of life and limb salvage rates.

Keywords

aortoiliac occlusive disease, pyoderma gangrenosum-like lesions, skin ulcers

INTRODUCTION

Aortoiliac occlusive disease (AIOD) is a segmental non-inflammatory obstructive vasculopathy commonly occurring in the infrarenal aorta and iliac arteries. It affects mainly patients with advanced atherosclerosis, diabetes mellitus, or hypercholesterolemia, and those with other risk factors such as cigarette smoking, age, family history, race etc.[1] The condition occurs in both sexes and people of all races, but is more common in the elderly, since the higher incidence of atherosclerosis of the lower extremities is associated with an increase in the general population's life expectancy.[2] As far as the occlusive atherosclerotic process is chronic, it leads to development of collateral blood vessels supplying the affected extremities, hence the clinical tissue ischemia and the cutaneous injury features may occur in different interval and severity.[3,4] It has been reported that the coexistence of AIOD, abdominal aortic aneurysm, ischemic heart disease, and cerebrovascular disease carry a very poor prognosis.[5] Based on the morphology and the level of lesions, the Trans-Atlantic Inter-Society Consensus (TASC) guidelines classified aortoiliac atherosclerotic disease TASC II C and D types and the standard recommended treatment is bilateral surgical bypass of the femoral arteries.[6]

Cutaneous manifestation varied from nonspecific inflammatory lesions through ulcers to gangrene of low extremities and the diagnosis might be difficult.[3] The pa-

tients report a severe intermittent claudication, ischemic rest pain, or burning and pain in the cutaneous lesions.

We present two Caucasian patients with similar medical histories in whom we suspected severe obstruction of the infrarenal aorta and iliac arteries because of the clinical and cutaneous features, which was verified by contrast angiography and Doppler ultrasonography.

CASE REPORT

Case 1

A 55-year-old woman was admitted to the Department of Dermatology, Alexandrovska University Hospital in Sofia with pustular lesions on the abdomen, and painful multiple ulcers located on the postoperative inguinal scar, the buttocks, and internal part of the left thigh. Pathergy reaction was positive and the working diagnosis was pyoderma gangrenosum (PG). The patient had a history of allergic reactions to non-steroidal anti-inflammatory drugs (NSAID) and house dust mites and concomitant arterial hypertension, diabetes mellitus, and ischemic heart disease, including past myocardial infarction.

Physical examination revealed vesiculopustular skin lesion on the abdomen ***(Fig. 1)*** *and deep ulceration with well-defined borders on a postoperative scar on the left inguinal fold. Multiple ulcers with a necrotic bottom, irregular edges, and erythematous halo were located on the left buttocks and internal region of the left thigh. Purpuric lesions were observed on the skin of the lower limbs and the left foot was edematous with violaceous colour. The left thumb was necrotic, with bullous lesions on its back aspect. Palpation of the distal superficial femoral artery showed no pulsations. Complete blood count and routine blood tests were within normal ranges except for cholesterol levels - 5.64 mmol/l (up to 5.2 mmol/l) and glucose levels - 10.3 mmol/l (reference range 3.6–6.1 mmol/l) levels. The prothrombin time and some other coagulation function markers were also impaired. Histopathologic investigation of a skin biopsy showed abscess formation in the dermis and dense lymphocytic infiltration around blood vessels, with extravasation of erythrocytes* ***(Fig. 2)****. Contrast aortography showed chronic obstruction of the left iliac artery with development of collaterals* ***(Fig. 3)****. Doppler ultrasonography of the left common femoral artery showed decreased pressure (40 mm/Hg) in comparison with the right one.*

Methylprednisolone (1 mg/kg/24 h) was started for PG and the dose was reduced step-wise over the next 2 months and combined with local ozone therapy. Therapy resulted in healing of the ulcers on the inguinal fold and internal aspect of the thigh and allowed completion of the surgical treatment of AIOD. Thromboendarterectomy of the femoral artery and bypass of the left aorto-femoral artery with an 8-mm prosthesis was performed in the cardio-vascular clinic. At the same time, a necrectomy of left thumb was performed. Treatment continued with pentoxifylline and acenocoumarin to complete resolution of the ulcers. The concomitant diabetes, cholesterol, and hypertension were controlled with adequate medications.

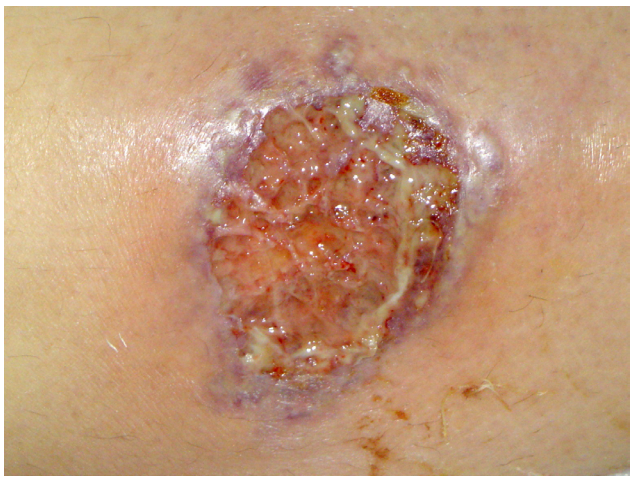

Figure 1. Pustular pyoderma gangrenosum-like ulceration on the abdomen of 55-year-old woman.

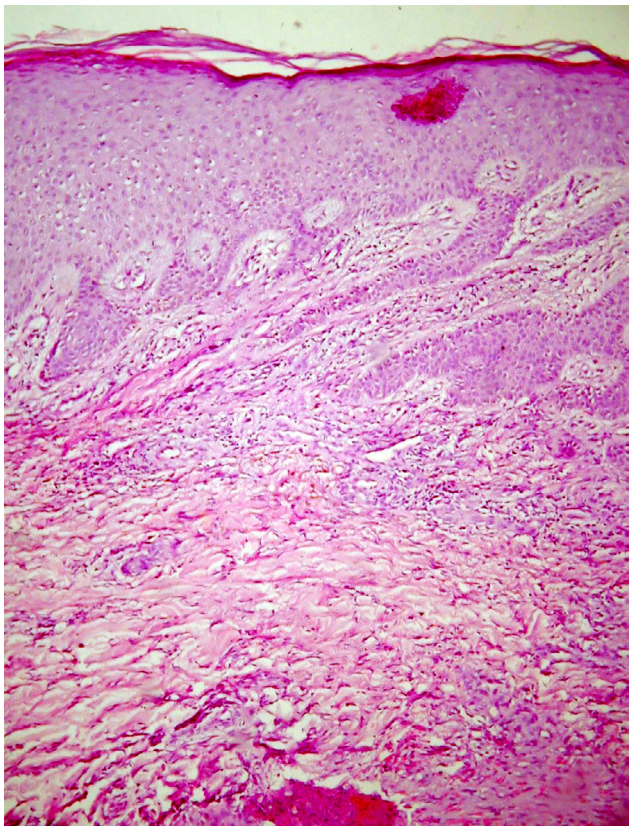

Figure 2. Histopathology of skin presenting acanthosis in epidermis, abscess formation and dense lymphocytic infiltration around blood vessels with extravasation of erythrocytes in dermis (H&E) ×200.

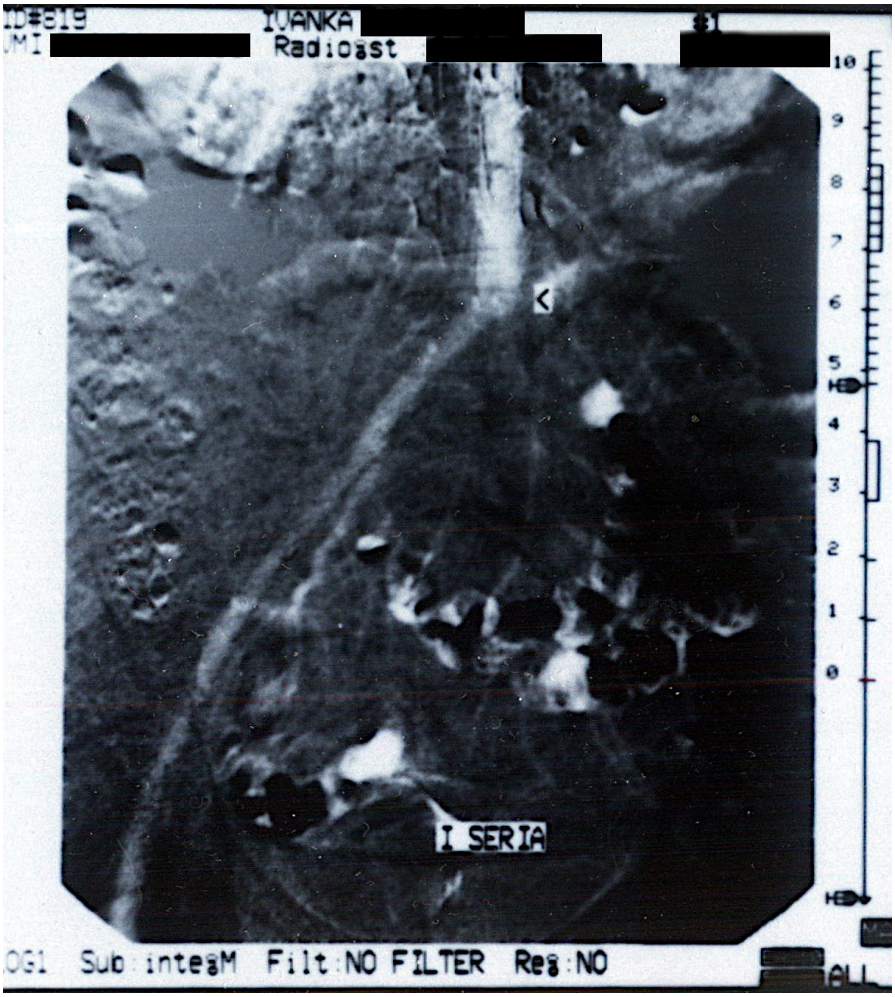

Figure 3. Contrast aortography showing occlusion of the left aorto-iliac artery.

Case 2

A 56-year-old male patient was admitted to our Department of Dermatology with a 3-year history of persistent swelling of both calves. He complained of formation of small ulcers followed by spontaneous re-epithelization; however, six months earlier, deep painful nonhealing ulcerations had appeared on the anterior aspect of his legs. The patient was a heavy smoker (>30 cigarettes/day). Previous treatment with diosmin and local re-epithelization agents was ineffective.

Physical examination revealed claudication after walking 15 feet and diffuse livedo reticularis, purpuric and pigmented maculae on both lower extremities. The patient's left leg was slightly thinner than the right. Four deep ulcerations, two around the left knee, one pretibial, and one around the left medial ankle ***(Fig. 4)*** *with callous borders and fibrotic bases* ***(Fig. 5)*** *were observed on the left leg. The arterial pulsations of the left tibial arteries were reduced but palpably detectable. No pulsations were found over the left femoral artery.*

Routine laboratory investigations were within the normal ranges except for the slightly elevated aspartate transaminase (46 U/l) and creatine (130 mmol/l). Doppler ultrasonography measured decreased pulsations of the left posterior tibial artery (40 mm/Hg) compared with the right (110 mm/Hg) and normal blood pressure (RR 130/90 mm/Hg) of the left brachial artery. Abdominal CT revealed homogenous hepato- and splenomegaly. Angiography found occlusion of the bifurcation of the left iliac artery and an enlarged, corkscrew lineal artery. The right femoral artery, however, was normally visualised due to collateral arteries. One month later, the obstruction of the iliac artery was surgically removed. Maintenance therapy was pentoxifylline, verapamil, and acetylsalicylate with local iodine povidone dressings.

DISCUSSION

Aortoiliac occlusive disease is large-vessel vasculopathy characterised by degenerated stenotic atheromatous obstruction, primarily affecting the infrarenal aorta and iliac arteries.[7] Obstruction may appear in the infrarenal aorta, common iliac, internal iliac, external iliac, or combinations of any or all of these vessels. Patients' complaints are variable, and patients may initially have no symptoms. In some

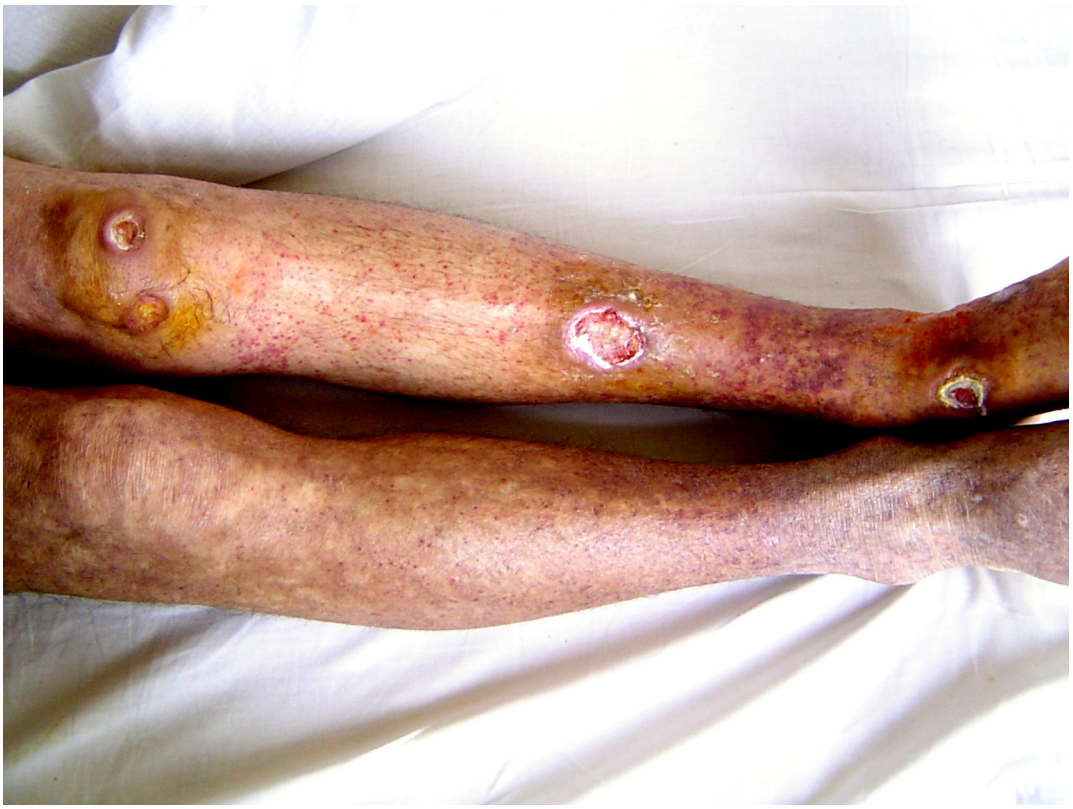

Figure 4. Arterial ulcers on the left leg, petechiae and reticular cyanotic erythema of the skin in in 56-year-old male.

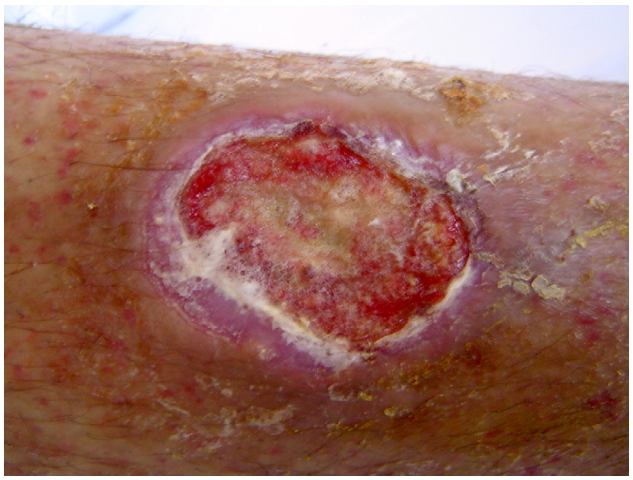

Figure 5. Ulceration on anterior aspect of the left leg with callous borders and a fibrotic base.

cases, non-specific symptoms, such as stiffness, paresthesia, reduced sensitivity or pain, and claudication appear. [8] Intermittent claudication, defined as pain or fatigue in a muscle or muscle group on repetitive use, is common. The anatomical level of claudication is significant, and when aorto-iliac artery is obstructed, pain occurs first in the hip or thighs.[8] Ischemic rest pain indicates an advanced stage of the disease. Neurogenic claudication is characterized by sensory symptoms that appear during exercise or while maintaining a fixed posture, and some patients may have bowel and bladder disturbance.[9]

Claudication in our patients was an alarming sign. The constellation of intermittent claudication, erectile dysfunction, and absent femoral pulses in men is termed Leriche syndrome (LS), named after the surgeon who first described the condition in 1923.[10,11] LS occurs when either pre-occlusive stenosis or complete occlusion of the infrarenal aorta is present. Our second patient had been a smoker for a long time and had all signs of LS – intermittent claudication, erectile dysfunction and absent femoral pulse. Murphy et al.[9] reported a 66-year-old-woman with AOID, who presented with symptoms of paresthesia from the buttocks to the thigh, and intermittent loss of bladder and bowel function associated with absence of femoral pulses and symptoms of claudication. Other diseases with obstruction of the abdominal aorta and its main branches are Takayasu's disease and Wegener's granulomatosis, two chronic segmental vasculitides of large arteries with unknown etiology.[12,13]

Our first patient presented with vesiculopustular lesions on the abdomen and postoperative cicatrix compatible with PG. Pyoderma gangrenosum following surgery has been repeatedly reported.[14,15] In this case surgery was carried out without risk of complications and with the use of subcutaneous sutures and systemic steroid cover.[16] Esnault et al.[15] reported a 68-year-old man with two recurring postoperative PG ulcers after two femoral bypasses. These cases

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illustrate the importance of precise differential diagnosis in non-healing wounds. It is also important to consider the rare possibility of underlying AIOD in patients with PG, and the preoperative workup is essential to prevent post-operative complications.

The evolution of the disease in our patients showed some characteristic cutaneous findings: (a) ulcerations and pustules sometimes resembling PG; (b) multiple deep and painful ulcers with callous borders and necrotic or fibrinous bottom, along the lower extremities; (c) petechial and purpuric lesions; and (d) livedo reticularis. All of these cutaneous findings were associated with angiography and Doppler ultrasonography, confirming arterial occlusion; aorto-iliac arterial reconstruction significantly promoted wound healing.

AOID should be distinguished from other low limb ischemic disorders such as peripheral arterial occlusive disease, superficial femoral artery occlusive disease, chronic lower extremity ischemia, peripheral emboli, and lower extremity atheromatous emboli syndrome.[17,18]

CONCLUSIONS

Collaboration between the dermatologist and vascular surgeon can shorten the diagnostic process and might be essential for ensuring optimal management of patients with aorto-iliac occlusive disease. Early determination of the diagnosis and surgical reconstruction of vessels should improve both the patient's quality of life and limb salvage rates. Patients need to be given education on lifestyle modification and risk to prevent progression of the disease, including smoking cessation, increased physical activity, and diet modification.

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Кожные проявления аорто-подвздошной окклюзионной болезни: два случая и обзор литературы

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Резюме

Аорто-подвздошная окклюзионная болезнь (АПОБ) представляет собой невоспалительную обструктивную васкулопатию, обычно поражающую пациентов с выраженным атеросклерозом, сахарным диабетом или повышенным уровнем холестерина, а также субъектов с другими факторами риска, такими как курение сигарет.

Сообщается о двух пациентах европеоидной расы (55-летняя женщина и 56-летний мужчина) с кожными язвенными поражениями АПОБ. В обоих случаях история болезни включает начальную хромоту нижних конечностей, множественные болезненные язвы вдоль ног и отсутствие пульса на поверхностных бедренных артериях. Тяжёлая обструкция как инфраренальной аорты, так и подвздошных артерий слева была продемонстрирована с помощью контрастной ангиографии и допплерографии.

Эволюция заболевания показала некоторые характерные признаки, в том числе язвы, подобные гангренозной пиодермии, как начальное кожное проявление АПОБ, множественные болезненные язвы вдоль нижних конечностей и аорто-подвздошную окклюзионную болезнь из-за атеросклероза.

Ранняя диагностика и хирургическая реконструкция сосудов у пациентов с АПОБ улучшали качество жизни и показатели сохранения конечностей.

Ключевые слова

аорто-подвздошная окклюзионная болезнь, поражения, подобные гангренозной пиодермии, кожные язвы