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CASE REPORT

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Isolated thigh ischemia in the course of iliofemoral arterial obstruction

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Abstract

A 72-year-old female presented rapidly developing ischemia of the skin and subcutaneous tissue of the antero--medial aspect of a thigh. An atypical clinical manifestation of ischemia led to delayed diagnosis and treatment that resulted in the development of large tissue loss in the anteromedial aspect of the thigh. Endovascular recanalization of recurrent occlusion in the ilio-femoral segment with rotational thrombectomy together with a multi-step local surgical treatment resulted in healing of the lesion.

Key words: chronic limb-threatening ischemia; thigh necrosis; thigh ischemia; percutaneous rotational thrombectomy

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Introduction

Chronic Limb Threatening Ischaemia (CLTI) most commonly results from the progression of atherosclerotic occlusive lesions in the arteries that supply blood to the limb [1]. The estimated incidence of the disease is 500–1000 per one million individuals per year [2]. The most important risk factors are smoking, older age, diabetes, and dyslipidemia [3, 4]. The disease most commonly manifests with rest pain followed by ulcers and gangrene. Once these lesions occur limb loss is imminent unless promptly revascularized [5]. Typically, the rest pain and gangrene affect the distal parts of the limb such as the toes, forefoot, or heel [6]. With a prolonged duration of ischemia, the lesions extend to the more proximal and deeper structures [7]. The purpose of this paper is to present a case of an atypical clinical presentation of recurrent CLTI with rest pain and gangrenous lesions affecting only the anterior surface of the thigh that was successfully treated with advanced endovascular techniques.

Case report

A seventy-two-year-old woman presented to a vascular surgery department because of severe pain in an anterior aspect of her right thigh. Seven months earlier because of an exacerbation of chronic right limb ischemia manifested by a short distance intermittent claudication and rest pain at night, she had undergone an endarterectomy of the external iliac artery (EIA) and common femoral artery (CFA) through a femoral approach. Due to proximal residual stenosis in the common iliac artery (CIA), a balloon expandable stent (9 × 58 mm) had been implanted. At that time superficial femoral artery (SFA), deep femoral (DFA), and popliteal artery (PA) were patent without any significant stenosis. Both surgery and postoperative course were uneventful, and cessation of the patient's complaints

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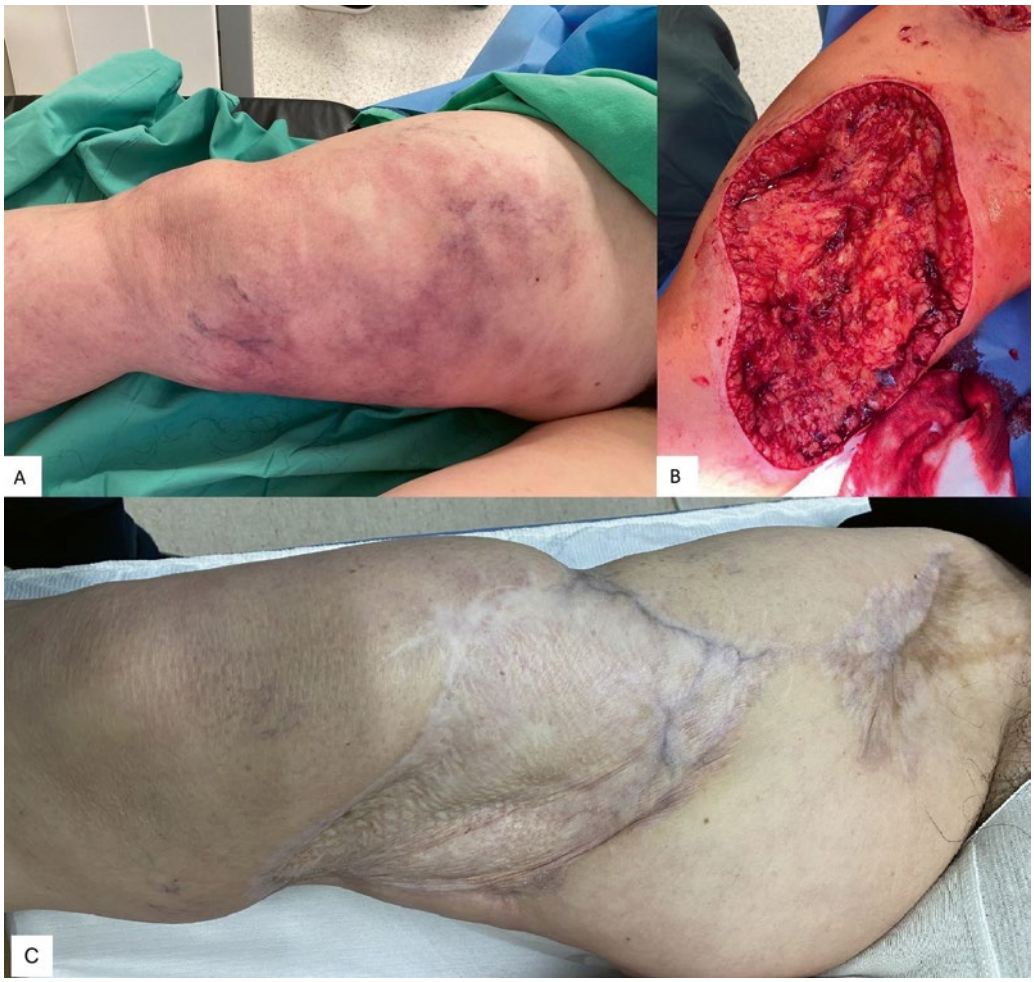

Figure 1. A. Cyanosis and edema of the anteromedial surface of the right thigh directly before surgical excision. **B.** Wound of the anteromedial surface of the right thigh directly after surgical excision. **C.** Completely healed thigh

was accomplished. Clopidogrel 75 mg and ASA 75 mg once daily and walking exercises along with cessation of smoking were recommended. Her past medical history included also percutaneous coronary intervention with drug-eluting stent implantation 10 years earlier and treatment for hypothyroidism. The patient presented typical vascular risks factors such as smoking, obesity, and hypertension.

On physical examination, tenderness, edema, and slight cyanosis on the anterior aspect of the right thigh were found. The temperature of the overlying skin was normal. The distal part of the limb was well perfused with a capillary refilling time below 2 seconds. Assuming that the patient's complaints did not result from ischemia anti-inflammatory and analgesic medications were recommended. The patient returned ten days later because of unremitting pain and extension of edema and cyanosis of the right thigh (Fig. 1A). The patient was afebrile and in a good general state. Angiography showed good patency of CIA, CIA stent, and internal iliac artery (IIA). EIA, CFA, and proximal segments of DFA and SFA were tightly stenosed (Fig. 2A). Distal parts of DFA, SFA, and PA and their branches were patent without any significant stenosis (Fig. 2B). Apart from increased concentration of C-reactive protein to 41,1 mg/l and mild anemia with hemoglobin of 10,7 g/dl, the results of laboratory tests were including serum creatinine, leucocytes, and procalcitonin were within normal limits. Percutaneous balloon angioplasty of EIA, CFA, and proximal segments of DFA and SFA with simultaneous excision of the affected area of the right thigh and groin was performed. The affected tissue had the appearance of skin and fat necrosis and involved only the skin and subcutaneous tissue without fascia and muscle involvement (Fig. 1B). Samples of excised tissue were sent for culture and intravenous treatment with clindamycin and ceftriaxone was started. On the 13th postoperative day, after the preparation with vacuum

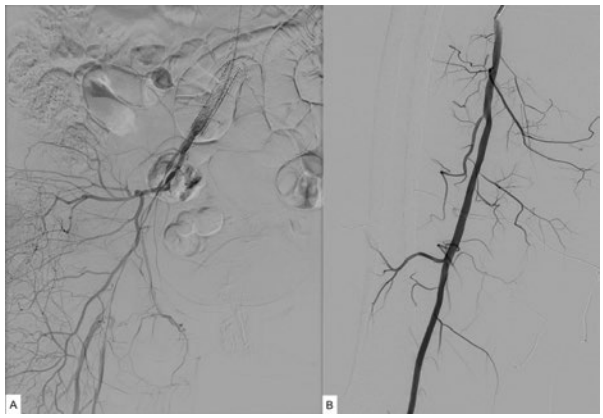

Figure 2. A. Intraprocedural arteriography showing patent common iliac and internal iliac arteries and tight stenosis of external iliac, common femoral, and proximal segments of deep femoral, and superficial femoral arteries. **B.** Distal parts of deep femoral and superficial femoral arteries and popliteal artery and their branches were patent without any significant stenosis

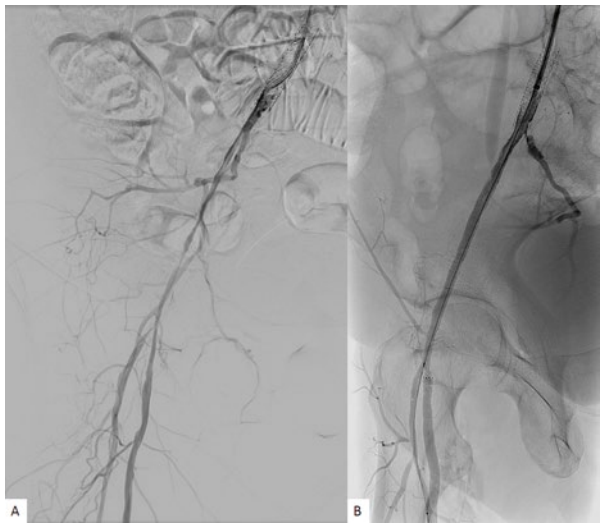

Figure 3. A. Intraprocedural arteriography showing critical restenosis of external iliac, common femoral, and deep femoral arteries. **B.** Intraprocedural arteriography after percutaneous rotational thrombectomy and superficial femoral artery stent implantation

therapy, the wound was covered with a split-thickness skin graft that healed the wound. Wound cultures were negative. The tissue culture for Actinomyces was also negative. The histopathology examination revealed the presence of foci of skin and fatty tissue necrosis with the formation of micro-abscesses.

Four months later the patient was admitted again because of the severe pain, necrosis lesions, and episodes of bleeding from the previously healed ulcer site. CT angiography showed again patent CIA and IIA, critically stenosed EIA, CFA, and proximal segments of DFA and SFA. Distal parts of DFA, SFA, and PA and their branches were patent without any significant stenosis. The wound was debrided, and samples were sent for microbiological examination. The culture revealed polymicrobial flora: *Enterobacter Cloacae ssp.*, *Staphylococcus haemolyticus* MRCNS, and *Staphylococcus epidermidis*. Despite repeated debridement and culture-guided intravenous antibiotic therapy the lesions were not healing. Taking into account the early recurrence of stenosis it was assumed that it was due to either thrombosis or intimal hyperplasia. Because of the tissue scaring after the previous procedures and the ulceration in the groin, it was decided to treat the patient endovascularly (Fig. 3A). Through a left axillary artery, a percutaneous rotational thrombectomy of right EIA, CFA, and proximal segments of DFA and SFA with Rotarex® S 8 Fr (Straub Medical) was performed. Due to a suboptimal result of thrombectomy an 8 mm self-expanding stent (Epic, Boston Scientific) was implanted in the EIA and a 6 mm self-expanding stent (Zilver Flex, Cook) was implanted in the proximal portion of SFA (Fig. 3B).

After the revascularization and debridement, the healing of the lesion was observed. Four months after the percutaneous thrombectomy the ulcer healed completely. During the 24-month follow-up, the patient remains asymptomatic without recurrence of the thigh ulceration (Fig. 1C).

Discussion

This paper reports a case of an atypical presentation of CLTI. The clinical signs of CLTI such as ulcers and gangrene most frequently affect the distal parts of the limb and penetrate to the deeper layers. In the described case the lesions involved only the skin and subcutaneous tissue of the anterior aspect of the thigh with uncompromised perfusion of the muscle of the thigh and of the distal limb. Due to atypical localization, the ischemia was initially erroneously excluded as the responsible factor and other causes of necrosis of subcutaneous tissue such as necrotizing fasciitis, actinomycosis, Clostridial cellulitis or myonecrosis or cellulitis caused by other pathogens was initially considered.

Due to the good general condition of the patient and the absence of characteristic signs such as crepitations, foul-smelling discharge, and hemorrhagic bullae as well as negative cultures Clostridial infection was discarded in the first place [8]. Actinomycosis, a disease caused by the anaerobic bacteria Actinomyces Israeli could have also given an observed clinical picture. The infection spreads by continuity provoking an inflammatory process with the formation of fistulae and abscesses discharging thick-yellow exudate. In the presented case a yellow exudate was absent and the Actinomyces

Israeli was not cultured. The inflammation of the skin and subcutaneous tissue could also have been caused by necrotizing fasciitis a polymicrobial infection of fascia and muscles characterized by rapidly progressing deterioration [9]. In our patients, the clinical progression was rather slow and neither fascia nor muscles were involved. Moreover, the culture of samples taken at the beginning of the disease was negative. Cellulitis due to other pathogens, mainly *Staphylococcus aureus* and *Streptococcus pyogenes*, was also considered [10]. This disease causes typical signs of inflammation such as local heat, redness, swelling, and pain. It is frequently preceded by minor trauma or bite [11]. In the present case, there was no history of trauma or bite, local heat was absent and, to be said again, cultures were negative. Normal levels of leucocytes and procalcitonin also argued against infection as a cause of observed symptoms. Most of all the healing of the wound after the recanalization of the external iliac artery and the recurrence of ulceration after its restenosis supports the main role of ischemia in the development of observed lesions. Between the 13th day and the 4th month after the index procedure, the patient was free of complaints which probably correspond to the time required for restenosis to develop and become clinically apparent.

The skin of the anteromedial surface of the thigh is perfused by the perforating arteries taking off mainly from SFA and DFA that were patent in the described case [12]. Moreover, the flow in these arteries as well as in the popliteal and tibial arteries must have been sufficient to maintain a satisfactory blood supply to the distal part of the limb. It could have been supposed that the perforating arteries had been injured during the previous endarterectomy. These arteries however whose number may vary from 8 to 14 are localized mainly in the mid-thigh region where the SFA was not exposed [13]. Moreover some of these arteries, so-called musculocutaneous perforators, supply blood also to the thigh muscles that did not suffer from ischemia in the presented case [14]. Also in arteriography, all branches of DFA and SFA were patent. The most probable explanation for observed ischemic lesions was a decrease in skin perfusion induced by compromised inflow. Clinical improvement after the restoration of inflow with subsequent deterioration with restenosis and ultimate healing after successful treatment of restenosis seem to confirm this hypothesis. Thus, occlusion of the external iliac and common femoral arteries may have an atypical clinical presentation in the form of progressive ischemia of the anterior surface of the thigh.

In a case of a subacute occlusion of the EIA surgical thrombendarterectomy or iliofemoral prosthetic graft would be probably the best and the most durable solution. However, because of the presence of the ulceration in the groin region, these procedures would be associated with a great risk of complications such as impaired wound healing or graft infection which may have disastrous consequences. Due to the subacute nature of arterial obstruction catheter-directed thrombolysis would have been unsuccessful and balloon angioplasty with stent implantation would have been associated with a risk of peripheral embolization. Percutaneous rotational thrombectomy is based on the action of a helical Archimedes screw that enables detachment of the occluding material, its aspiration and fragmentation, and transportation of the material out of the patient's body [15]. The use of percutaneous rotational thrombectomy removed almost completely the occluding lesion and allowed for safe and effective balloon angioplasty with stent implantation [16].

Conclusions

Chronic limb-threatening ischemia may have an atypical clinical manifestation with isolated involvement of the skin of the thigh and absence of distal limb ischemia. Percutaneous rotational thrombectomy is a safe and effective method of treatment of subacute arterial occlusion.

Conflict of interest

None.

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