

Case Report

Acroangiokeratitis- An Outcome of Pregnancy Induced Stasis? -A Case Report

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ABSTRACT

Acroangiokeratitis is a benign vascular disorder with variable presentation. It is usually associated with an underlying arteriovenous malformation (AVM) or chronic venous insufficiency. A rare case of a 35-year-old female, a defaulter to tuberculosis treatment, with recurrent painful ulcers over her left leg post-pregnancy without any evidence of underlying AVM is presented here.

Keywords: Acroangiokeratitis of Mali, Arteriovenous malformation, Dermite ocre of Favre, Pseudo-Kaposi sarcoma, Stasis dermatitis

INTRODUCTION

Acroangiokeratitis is a rare, benign condition which begins as violaceous macules and patches, gradually evolving into soft, non-tender red-to-purple papules, nodules or indurated plaques with oedema over the lower limbs.^[1] It represents a reactive angiodysplasia of cutaneous blood vessels.^[2] Although the precise aetiology of acroangiokeratitis remains unclear, venous insufficiency is identified as a common underlying mechanism in most cases.^[1-3]

Acroangiokeratitis has two types: Mali type, linked to chronic venous insufficiency (CVI), usually seen in the elderly and Stewart-Bluefarb type, associated with arteriovenous malformations (AVMs). Upper-limb involvement with ulceration suggests AVMs, while lower-limb cases are typically due to venous insufficiency. Acroangiokeratitis can present as Dermite ocre of Favre in pregnancy, post-arteriovenous shunt for haemodialysis and in post-thrombotic syndrome, lower-limb paralysis, Prader-Labhart-Willi syndrome, Klippel-Trenaunay-Weber syndrome and stump dermatosis in amputees.^[1,3,4]

CASE REPORT

A 35-year-old otherwise healthy woman, hailing from a rural region presented with a chronic painful ulcer on her left leg for 1.5 years and a history of incomplete tuberculosis treatment 10 years ago. The patient, G2P2L2A0, had a normal vaginal delivery 15 years ago and lower segment caesarean section 4 years ago. She was asymptomatic until 1-week postpartum after her first delivery, when she developed purplish-black discolouration, painful swelling and oozing lesions on her left foot and ankle, spreading bilaterally to the legs and forearms within 15 days. The lesions persisted for 1.5 months, requiring surgical intervention.

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Similar lesions recurred 1.5 years after her second pregnancy, evolving into painful, reddish-to-purplish raw areas over the left leg for the past year. Examination revealed a 4.5 × 5 cm ulcer with sloping edges, erythematous base and surrounding hyperpigmentation on the left leg, along with a hyperpigmented plaque over the left malleolus. Multiple scars with hyperpigmentation were seen on the bilateral legs and forearms, with soft, non-tender swellings on the right forearm and left leg and intact arterial pulsations [Figure 1a and b]. No palmar or plantar changes, palpable thrill or bruit was present.

Acroangiodermatitis, lupus vulgaris, stasis dermatitis and angiosarcoma were kept as differentials. Routine investigations were normal, human immunodeficiency virus negative, Mantoux positive and sputum acid fast *bacilli* (AFB) and cartridge based nucleic acid amplification test (CBNAAT) negative for tuberculosis.

Colour Doppler study ruled out venous insufficiency and AVM. Histopathological examination revealed epidermal hyperplasia with flattened rete ridges and compact hyperkeratosis. There was a mild superficial perivascular lymphohistiocytic infiltrate in the papillary dermis, along with a lobular pattern of superficial and deep neo-lymphovascular proliferation. Haemosiderin deposits, extravasated red blood cells and thickened, hyalinised collagen were seen in deep dermis [Figure 2a and b]. Based on the histopathological examination, the diagnosis of acroangiodermatitis of Mali was made. The patient was started on oral and topical antibiotics along with regular clean and meticulous wound care. She was then referred to general surgery whereby split-thickness skin grafting was advised.



Figure 1: (a) A solitary ulcer, measuring 4.5 × 5 cm², with well-defined margins, sloping edges and an erythematous base, surrounded by hyperpigmented skin, located on the posterior aspect of the left leg. (b) A single well-to-ill-defined, skin-coloured to hyperpigmented plaque with overlying scaling over the left lateral malleolus.

DISCUSSION

When angiodermatitis occurs with ulceration on the hand, it strongly suggests the presence of a shunt, whereas on the lower leg is frequently associated with CVI or paralysis. AVMs begin as faint macular erythema, expanding post-puberty or after trauma, later developing a violaceous hue, mass, warmth, thrill, bruit and potential ulceration or bleeding.^[2,4] Acroangiodermatitis in pregnancy, known as 'Dermite ocre of Favre', presents as purpuric macules and plaques over venous varicosities, often with oedema, sclerosis and ulceration, reflecting CVI. The lesions may progress to the dorsa of the feet and toes, persisting for months or years, supporting the case's chronicity and pathogenesis.^[5]

Most cases of acroangiodermatitis are linked to signs of venous insufficiency; however, Rao *et al.* found no evidence of venous insufficiency in 9 out of 10 cases.^[6] In our case, Doppler studies were normal and no underlying venous insufficiency or AVM was demonstrated. Notably, Hung *et al.* reported a similar case in a young woman without underlying venous insufficiency and hence termed it as 'spontaneous'.^[3] In addition, rare cases without venous dysfunction have been documented, such as in the study by Coban *et al.*^[2]

Acroangiodermatitis, a variant of stasis dermatitis resulting from capillary hypertension, chronic hypoxia and angiogenic stimulation, leads to fibroblast proliferation and

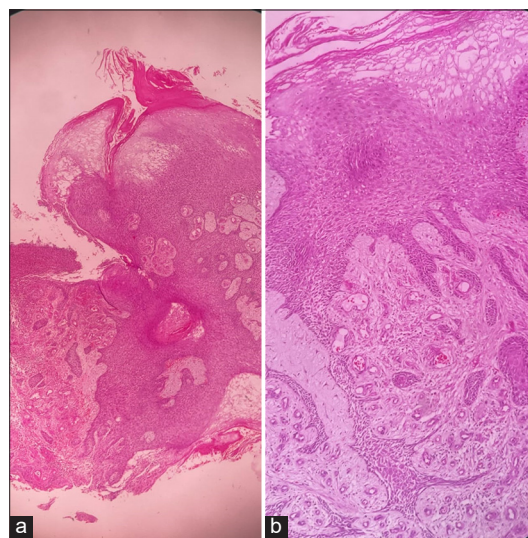


Figure 2: (a) Microphotograph showing epidermal hyperplasia, compact hyperkeratosis, neo-lymphovascular proliferation, and dermal changes including haemosiderin deposits, extravasated RBCs, and hyalinized collagen (H&E, x10); (b) Microphotograph showing lobular pattern of neo-lymphovascular proliferation in both superficial and deep dermis, haemosiderin deposition, extravasated RBCs and collagen deposition (H&E, x40). H&E: Haematoxylin and eosin.

neovascularisation.^[2] While most cases are associated with venous insufficiency, rare cases occur without it, as seen in this patient. Doppler studies were normal, ruling out CVI and AVM, aligning with previous reports of 'spontaneous' acroangiodermatitis.

Treating acroangiodermatitis is crucial to prevent complications such as infections bone demineralisation, soft tissue destruction and congestive heart failure. Management focuses on treating underlying vascular disorders, providing wound care and considering surgical interventions wherever necessary.^[1,2]

CONCLUSION

Uncomplicated or classic acroangiodermatitis is somewhat easier to recognise and diagnose. However, the spontaneous onset of acroangiodermatitis following pregnancy, as observed in this case, is a rare and underreported clinical entity, making it prone to being overlooked or misdiagnosed. This underscores the need for increased awareness and research, as delayed diagnosis can lead to complications and undue chronicity can further worsen the quality of life.

Ethical approval: Institutional Review Board approval is not required.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts

will be made to conceal their identity, but anonymity cannot be guaranteed.

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