Reticulate erythema with ulceration

Report of a Case

A 60-year-old woman presented to the dermatology outpatient department of the Postgraduate Institute of Medical Education and Research, Chandigarh with a painful, erythematous lesion of 1-year duration on the right buttock. There were superficial ulcerations at two places for 2 months. The lesions had not responded to multiple therapeutic agents, including oral antibiotics and topical corticosteroid cream. Her medical history was not significant for any systemic disease including underlying thromboembolic events and she had never smoked.

On examination, lacy, nonblanchable, reticular erythema diffusely involving the right buttock was seen. Two ulcers of size 5 cm × 3 cm and 2 cm × 2 cm covered with hemorrhagic crust were present [Figure 1]. There was no local raise in temperature or any underlying bruit. Femoral, popliteal, posterior tibial and dorsalis pedis pulsations were palpable on both sides. No gangrene or clinical signs of chronic venous insufficiency were observed. There was no lymphadenopathy. A skin punch biopsy was obtained and submitted for histopathological examination [Figure 2a and b].

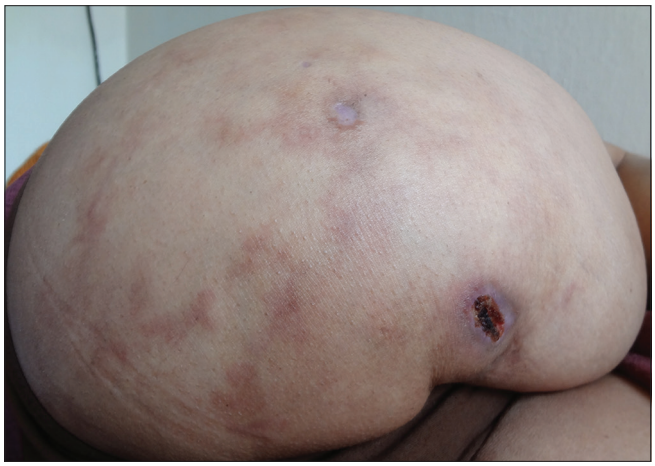

Figure 1: Lacy reticular erythema diffusely involving the right buttock. Two ulcers of size 5 cm × 3 cm and 2 cm × 2 cm covered by hemorrhagic crust

Investigations and Follow-up

Biopsy showed diffuse proliferation of CD31 positive endothelial cells throughout the dermis [Figure 2]. Direct immunofluorescence was negative. Serum antinuclear antibody and antiphospholipid antibody were negative. Coagulogram, kidney function tests and serum lipid profile were normal. Computed tomographic angiography of abdominal aorta and lower limb vessels revealed short segment partial thrombosis of the anterior division of right internal iliac artery.

Question

What is the diagnosis?

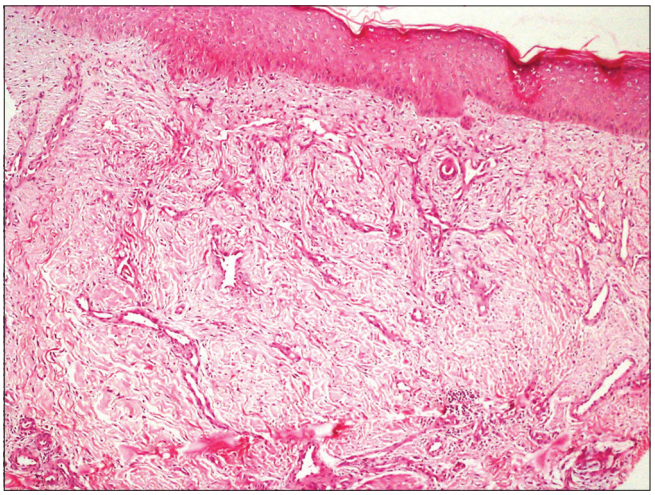

Figure 2a: Photomicrograph showing epidermis with flattened rete pegs, with proliferating and anastomosing small vessels in the dermis (H and E, ×100)

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Quick Response Code:	Website: www.ijdvl.com
	DOI: 10.4103/ijdvl.IJDVL_634_16

How to cite this article: Bhattacharjee R, Vinay K, De D, Sinha A, Saikia UN. Reticulate erythema with ulceration. Indian J Dermatol Venereol Leprol 2017;83:622-4.

Received: July, 2016. **Accepted:** December, 2016.

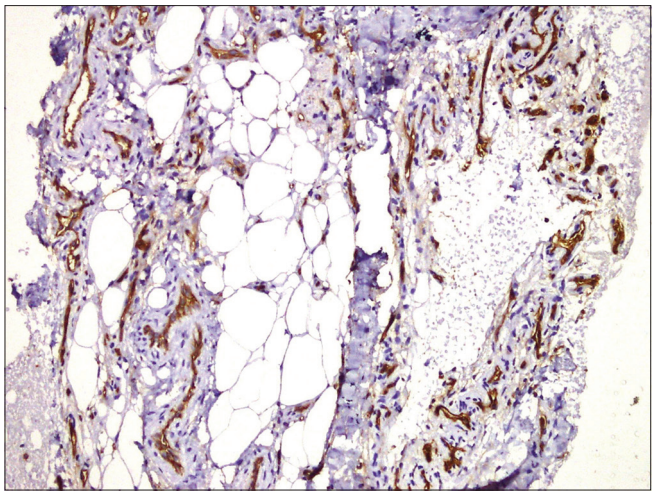

Figure 2b: Photomicrograph showing CD31 positivity in the endothelial cells of the vessels (IHC, ×200)

Answer

Diffuse dermal angiomatosis.

Our patient was treated with oral pentoxifylline 800 mg thrice daily and within days, there was significant improvement in pain. At 8 weeks follow-up, the skin lesions had improved considerably, the ulcers had healed and pain had resolved. Pentoxifylline was tapered and stopped by 16 weeks. At 18 months follow-up, she continues to be in clinical remission.

Discussion

Reactive angiomatosis are a rare set of angioproliferative disorders grouped together under a unifying term cutaneous reactive angiomatosis.[1] Clinically, the disorders are indistinguishable and present with similar features as erythematous, violaceous purpuric plaques or reticulate erythema with or without ulceration. Diffuse dermal angiomatosis, a subtype of cutaneous reactive angiomatosis has been classically described in association with atherosclerotic occlusion of the peripheral arteries.[2,3] The buttock, however, is an uncommonly reported site.

The pathogenesis of cutaneous reactive angiomatosis in general and diffuse dermal angiomatosis in particular is largely unknown. Various authors have suggested hypoxia as a central feature in the pathogenesis of diffuse dermal angiomatosis.[1-3] Its occurrence in patients with severe atherosclerosis and thrombotic or embolic occlusion of vessels further supports this theory.[4] It is speculated that hypoxia-induced expression of vascular endothelial growth factor and other cytokines stimulate the proliferation of endothelial cells.

The differential diagnosis of diffuse dermal angiomatosis includes vasculopathy, medium to large vessel vasculitis, acroangiodermatitis and benign or malignant vascular tumor. Histopathology helps differentiate diffuse dermal angiomatosis from other clinical differentials with similar presentation. Diffuse dermal angiomatosis is characterized by a diffuse proliferation of endothelial cells interstitially arranged between the collagen bundles within the full thickness of the dermis. In some areas, the proliferating cells may have a spindle-shaped appearance and a vacuolated cytoplasm forming small vascular channels.[1] Immunohistochemistry with CD31 and CD34 is useful in confirming the normal endothelial phenotype of cells lining the vessels.

We were unable to find a uniformly effective treatment of this dermatosis in literature. The management requires improving underlying tissue hypoxia and ischemia. Smoking cessation and a strict control of cardiovascular risk factors are imperative. Pentoxifylline use in our case was guided by the fact that it affects almost all factors responsible for blood viscosity.[5]

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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