Skin-Colored Papules, Nodules, and Bullae on the Shins in an Obese Female

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A 61-year-old obese, otherwise healthy, woman presented with asymptomatic swelling on both feet and shins along with mildly pruritic papulonodules and vesicles of gradual onset with multiple new lesions since 4 years. The vesicles used to coalesce and rupture, discharging clear fluid, and heal spontaneously. There were no similar complaints in family members. On examination, she was obese (BMI was 36 kg/m^2). Translucent papulonodules, few tense vesicles, and depigmented patches with perifollicular pigmentation along with pitting edema were noted on the shins [Figure 1, inset]. The feet were spared of lesions, and Stemmer's sign was negative. There was no clubbing, hyperhidrosis, hypertrichosis, thyroid swelling, or exophthalmos. Venous insufficiency was not noted clinically and on Doppler ultrasound. The thyroid profile including autoantibody screen and diethylcarbamazine provocation test for filariasis was negative.

Histology from a nodule revealed epidermal flattening with attenuated rete ridges and subepidermal split. In the papillary and mid-dermis, vertically oriented blood vessels, dilated lymphatics, increased number of fibroblasts, and edema were observed [Figure 2a]. The subcutaneous tissue was normal; however, a dilated eccrine duct was noted [Figure 2b]. Mucinous infiltration (on Alcian blue stain) was evident [Figure 2c].

Question

What is your diagnosis?

Answer

Obesity-associated lymphedematous mucinosis (OALM)

Discussion

Obesity-associated lymphedematous mucinosis (OALM) is a rare, relatively recently described entity characterized by pretibial edema and mucin deposition in the absence of thyroid disease.[1] Clinically, it presents as skin-colored, translucent or erythematous, brown papules, nodules, and in cases of intense lymphedema, vesicles with underlying edema, and erythema characteristically localized to the shins.[1,2] Other sites such as the feet and thighs are almost universally spared. Fewer than 50 cases have been described. Obese middle-aged to elderly individuals are usually affected.

The commonly proposed pathomechanism is that obesity causes reduced tissue oxygenation and accumulation of proteinaceous lymphatic fluid due to lymphatic obstruction. The latter eventuates in accumulation of lymphatic fluid rich in fibrinogen, other coagulation factors, and albumin. All these factors culminate in increased mucin production by fibroblasts.[1]

Histologic findings include epidermal atrophy, papillary dermal mucin deposition, vertically oriented vessels in the superficial and mid-dermis, increased fibroblasts with variable fibrosis and occasional subepidermal split.[1,3] All these features were noted in our patient.

The major differential diagnosis is pretibial myxedema associated with Grave's disease. Clinically, the absence of other features of thyroid disease, hyperhidrosis, or hypertrichosis on the shins and normal thyroid function tests obviated this possibility. Histopathologic features of pretibial myxedema such as orthohyperkeratosis, acanthosis, reticular

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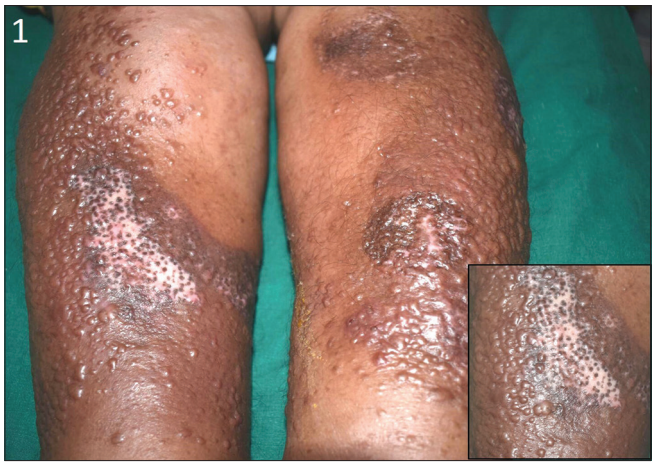

Figure 1: Translucent papulonodules and few tense vesicles and bullae on both shins. Depigmentation with perifollicular repigmentation probably as a consequence of healing of ruptured subepidermal bulla are noted (inset: close-up view)

dermal and subcutaneous mucin deposition, presence of mast cells, and absence of vertically oriented vessels are unlike OALM.[1,3]

Stasis mucinosis (due to venous insufficiency) is part of the OALM spectrum and a close differential but has histopathologic differences including mucin deposits around eccrine glands and hair follicles (unlike in our case).[1]

Other plausible differential diagnoses include epidermolysis bullosa pruriginosa, pretibial bullous pemphigoid, and filarial lymphedema. The absence of symptoms of filariasis, sparing of feet, absent Stemmer's sign, and negative diethylcarbamazine provocation test rule out filariasis, which usually presents unilaterally. Extensive dermal mucin deposits and paucity of inflammatory infiltrates discount the possibility of bullous pemphigoid. The absence of intense pruritus, family history, and nail dystrophy and the presence of dermal mucin makes epidermolysis bullosa pruriginosa unlikely. Clinically, the presence of translucent papulovesicles, absence of pain, tenderness, and deep-seated nodules and narrowing of the lower leg discounted the possibility of lipodermatosclerosis. Histopathologically, prominent mucin deposition, vertically oriented vessels, subepidermal bullae, absence of vascular plugging, erythrocyte extravasation, hemosiderin deposition, and panniculitis further discounted this possibility.

The peculiar healing pattern (hypopigmentation with perifollicular pigmentation), akin to subepidermal bullous dermatoses, has not yet been described in OALM. This probably resulted from bursting of subepidermal bullae in this case. The dilated eccrine duct [Figure 2b] has also not been noted previously. Presumably, edema and mucinous infiltration obstructed sweat flow through the duct. However, clinically, there was no history of hypohidrosis, suggesting that this finding is, perhaps, incidental.

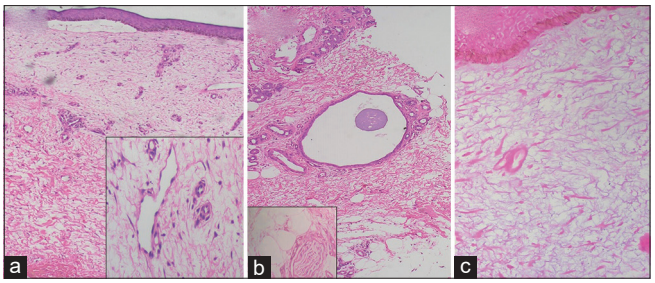

Figure 2: (a) Epidermal flattening, subepidermal split, dermal edema (maximum in the papillary dermis), and vertically oriented vessels in the upper dermis along with dilated lymphatics (hematoxylin and eosin, 5×). (Inset) edema, abundant fibroblasts, and a dilated lymphatic are seen (hematoxylin and eosin, 20×). (b) A dilated eccrine duct is seen in the lower dermis (hematoxylin and eosin, 5×) with normal subcutaneous fat (inset: hematoxylin and eosin, 20×). (c) Alcian blue staining (pH 2.5) confirms the presence of mucin (blue tinge) accompanying dermal edema (20×)

Therapy comprises of low caloric diet and compression garments, pentoxifylline, and topical or intralesional steroids.[2] Response to these modalities is generally partial. Unfortunately, our patient could not be followed up to assess treatment efficacy. Although we were unable to find similar such cases reported from the Indian subcontinent, considering the current "obesity epidemic," such cases may be seen more frequently in future, which mandates awareness of this rare, disfiguring entity.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/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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Conflicts of interest

There are no conflicts of interest.

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