Recurrent Pustular Erythema Nodosum Leprosum: A Rare Case Report

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Sir,

A 34-year-old male had been diagnosed as a case of lepromatous leprosy, based onthe presence of multiple skin-coloured to erythematous nodules with wrinkling on the surface over ear lobules, trunk and extremities and bilateral thickened and tender ulnar and common peroneal nerves. He was on multidrug multibacillary regimen since 3 months prior. There was history of episodes of reddish painful skin lesions with joint pain, for which he had received variable doses of oral prednisolone on and off for 1 month and the patient had stopped prednisolone on his own after lesions subsided.

He presented to our department, on two consecutive occasions, with multiple pustular lesions over erythematous tender nodules present mostly on the extensor aspects of limbs and trunk [Figure 1a and b]. Older lesions showed central ulceration with crusting. There were no other comorbidities and no similar family history. With a diagnosis of lepromatous leprosy with erythema necroticans with secondary impetinization, patient was admitted. Slit skin smear from the skin coloured nodules showed presence of acid-fast bacilli. Gram stain of the pustules showed only neutrophils. Culture sensitivity of the aspirated pus from the intact pustules did not grow any organism. Biopsy of the pustular lesion showed intraepidermal vesicle located in the sub-corneal region containing collection of neutrophils and histiocytes, with dermis showing collection of sheets of foamy histiocytic cells admixed with few polymorphs and infiltration of nerves and adenexal structure by these histiocytic cells [Figure 2a-d]. Fite Faraco stain revealed presence of numerous acid fast bacilli in the cytoplasm of the foamy histiocytes and endothelial cells of blood vessels [Figure 2e and f]. Other routine investigations such as complete blood count, liver and renal function tests and anti-retroviral tests were within normal

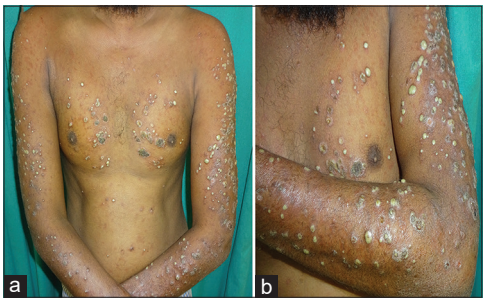

Figure 1: (a) Multiple pustules present over limbs and trunk. (b) close up view showing multiple pustules and erythematous papules. Few pustules have ruptured to form ulcers with crusting

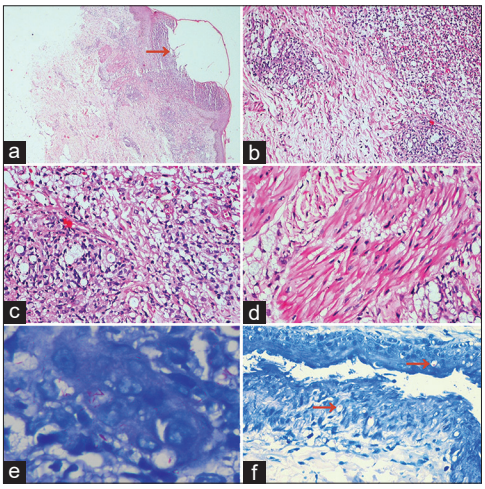

Figure 2: (a) Scanner view showing intracorneal cleft containing collection of neutrophils and histiocytes. [H and E, 40×]. (b) low power view of dermis showed collection of sheets of foamy histiocytic cells admixed with few polymorphs. [H and E, 100×]. (c) high power view showing collection of sheets of foamy histiocytic cells and few polymorphs in dermis. [H and E, 400×]. (d) high power view showing infiltration of histiocytes in arrector pilli. [H and E, 400×]. (e) presence of numerous acid fast bacilli in the cytoplasm of the foamy histiocytes. [Fite Faraco stain, oil immersion]. (f) presence of bacilli in the endothelial cells of the blood vessels. [Fite Faraco stain, oil immersion]

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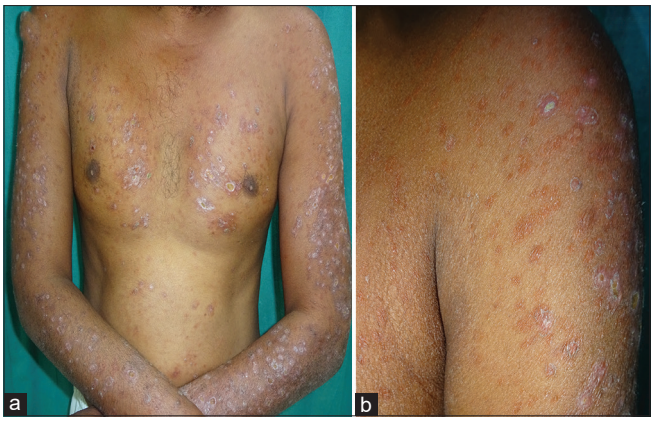

Figure 3: (a) Healed lesions over the trunk and extremities. (b) close up image of the healed lesions showing superficial scars

limits except low level of haemoglobin (8.9 gram/dL). A final diagnosis of lepromatous leprosy with pustular erythema nodosum leprosum (ENL) was made and patient was treated with oral prednisolone 60 mg, which was gradually tapered over next 2months with improvement of the lesions and multidrug therapy was continued [Figure 3a and b]. The second episode had precipitated on tapering the dose of prednisolone to 35 mg. The patient was hence started on thalidomide and oral steroid was tapered. Patient is on regular follow-up.

ENL may present with various atypical forms, pustular forms have been described rarely.[1,2]

Higher bacillary load and, in few instances, bactericidal drugs like ofloxacin, have been implicated in the pathogenesis of pustular form.[2] The presence of Lepra bacilli in the endothelial cells is not uncommon and is reported to induce production of anti-endothelial cell antibodies, which play a role in pathogenesis and dissemination of the disease, mainly in lepromatous forms.[3]

The differential diagnoses of pustular form of ENL can be secondary impetinization of ENL lesions and varicella. Detailed history of evolution of lesion and gram stain and culture sensitivity can differentiate from impetigo.Our patient presented with consecutive episodes of pustular forms of ENL.

This atypical variant can not only cause diagnostic dilemma, but can cause delay in appropriate treatment and pustular form being a severe form of type 2 lepra reaction, unless managed timely can lead to permanent damage of various organ systems.

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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Nil.

Conflicts of interest

There are no conflicts of interest.

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