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Case Report

Bullous erythema nodosum leprosum

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Introduction

Leprosy is a chronic, slowly progressive, granulomatous infection caused by *Mycobacterium leprae*. Reactions in leprosy, also known as lepra reaction, are not uncommon. They are of two types – Type 1 lepra reaction (occurring in borderline disease) and the Type 2 lepra reaction (occurring in lepromatous disease).¹ Skin lesions in lepra reaction generally manifest as exacerbation of existing skin lesions which become more erythematous and oedematous as well as appearance of fresh similar lesions (Type 1) or appearance of crops of numerous evanescent, erythematous, tender nodules and plaques (Type 2). Bullous lesions are a rarity in lepra reactions. Recently we came across a case of leprosy with Type 2 lepra reaction having bullous lesions which prompted us to report the case.

Case report

A 54 years old male, a known case of lepromatous leprosy on WHO multidrug therapy (MDT) for the past 18 months,

presented with acute onset of fever, malaise and body ache and multiple painful reddish swellings and fluid filled lesions in different parts of body of 12 days duration. He gave history suggestive of several earlier episodes of Type 2 lepra reactions with erythema nodosum leprosum (ENL) lesions which were managed with moderate to high doses of corticosteroids with improvement. However attempts to taper off the steroids were never successful. For the first time on this occasion he had also developed multiple fluid filled lesions. There was no history of having any precipitating factor for the lepra reaction.

Dermatological examination revealed multiple bilateral almost symmetrically distributed erythematous tender nodules and plaques on face, extremities and trunk. He also had multiple bullous lesions on trunk (Figs. 1 & 2). In the beginning the bullae were tense containing clear fluid but latter they became flaccid, and ruptured to form erosions and crusts. Nodules and plaques were evanescent and recurrent, individual lesions lasting for few days, later healing with hyper pigmentation. Nikolsky's sign and bulla spread sign were negative. Infiltrations were present over earlobes and in eyebrow region with supraciliary madarosis. Bilateral ulnar and common peroneal nerves were thickened uniformly but non-tender. There was no mucosal involvement. There was no feature suggestive of neuritis, iridocyclitis, orchitis or any other systemic involvement.

Investigations revealed polymorphonuclear leukocytosis and raised ESR. Biochemical investigations were normal. Slit skin smear examination showed fragmented acid fast bacilli with bacteriological index (BI) of 3+. Tzanck smear did not show any acantholytic cells. Bacteriological cultures from bullous fluid were sterile. Occasional fragmented AFB were seen in blister fluid. Skin biopsy from bullous lesion showed subepidermal bulla with diffuse polymorphonuclear cell infiltrate in

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Fig. 1 – Bullous lesions.

the dermis (Fig. 3) with a few foamy macrophages and histiocytes, perivascular inflammation was also present (Fig. 4).

He was managed with a course of systemic corticosteroid starting at 60 mg/day and tapered over a period of month, antileprosy drugs were continued. He showed good response to this treatment. Both bullous and nodular lesions started healing in about a week's time. He was started on thalidomide 200 mg twice daily for a month and later on 100 mg twice daily for 3 months and steroids were tapered off. There was no recurrence of the ENL lesions during the follow up period of about 6 months.

Fig. 2 – Multiple bullous lesions.

Fig. 3 – Heavy adnexal infiltrate H&E 10×.

Discussion

Lepra reactions reflect abrupt changes in the host parasite immunologic balance and are associated with acute clinical exacerbation.² Type 2 reaction is an immune complex reaction and is seen mostly in lepromatous (multibacillary) cases.³ It has been reported that over 50 percent of lepromatous leprosy patients and 25 percent of borderline lepromatous leprosy patients experience an erythema nodosum leprosum (ENL) reaction.² During Type 2 lepra reaction these antibodies combine with *M. leprae* antigen to form immune complexes which circulate and get deposited in various tissues, activate complement and damage these tissues.³ Though standard multidrug therapy of 2 (now 1) years kills most of the lepra bacilli, the elimination of dead lepra bacilli from the tissues is very slow and takes many years. Moreover the antibodies continue to react with the antigens even after completing antileprosy multidrug therapy. Erythema nodosum leprosum (ENL) is the typical lesion of Type 2 lepra reaction and consists of crops of numerous evanescent, erythematous, tender nodules and plaques over extremities, trunk, face and other parts of body. Due to immune complex mediated damage there can also be lymphadenitis, neuritis, iridocyclitis, arthritis, synovitis, myositis, epididymo-orchitis, glomerulonephritis etc. Bullous lesions in Type 2 reactions are very rare.

Fig. 4 – Leucocytoclasia H&E 40×.

There are only a few report of bullous reaction in leprosy.^{3–7} Bullous lesions in leprosy may be manifestations of severe ENL reaction in patients having very high bacillary load.³ Bullous eruptions have been reported during treatment with rifampicin⁸ and dapsone.⁹ Pustular, ulcerated, hemorrhagic and erythema multiforme-like lesions are some other uncommon reported presentation of ENL. This presentation should be differentiated from, bullous drug eruptions, pemphigus vulgaris, bullous pemphigoid and other blistering diseases. However sudden onset of disease, uninvolved oral mucosa, appearance of bullous lesions with classic ENL lesions, absence of acantholytic cells in Tzanck smear rule out these possibilities. Corticosteroids and thalidomide are well known drugs for the management of type 2 lepra reaction. However corticosteroids are associated with side effects of long-term use which can be minimized with the use of thalidomide.

The present case is interesting in view of rare presentation of bullous ENL reaction in a multibacillary leprosy case while on antileprosy multidrug therapy and for the good response to thalidomide in preventing relapses of ENL.

Conflicts of interest

All authors have none to declare.

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