CASE REPORT

Bullous erythema nodosum leprosum: a case report from Nepal

A. RIJAL*, S. AGRAWAL*, A. AGARWALLA* & M. LAKHEY**
*Department of Dermatology and Venereology, B.P. Koirala Institute of Health Sciences, Dharan, Nepal
**Department of Pathology, BP Koirala Institute of Health Sciences, Dharan, Nepal

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Summary A patient with lepromatous leprosy, while on WHO multidrug therapy (MDT) for multibacillary disease, was diagnosed as having dapsone syndrome with recurrent episodes of bullous lesions on the lower extremities for 4–5 years. The lesions were associated with high-grade fever. Examination revealed multiple hypopigmented macules on the limbs. Multiple atrophic scars were also found on the buttocks and lower limbs. Bilateral ulnar, radial cutaneous and lateral popliteal nerves were thickened. On day 10 of WHO-MB-MDT he developed a flaccid bulla on the lower leg. Skin slit smear showed a bacterial index (BI) of 3+ and the histopathology was consistent with type II reaction. High dose corticosteroid therapy was started but he continued to have new lesions, and was therefore referred to a centre where thalidomide was available. Clinical response was good and he remained symptom-free after gradual reduction in dosage. ENL should be differentiated from bullous drug reactions, pemphigus vulgaris, bullous pemphigoid and other blistering diseases.

Introduction

Erythema nodosum leprosum (ENL) is a common manifestation of type II reaction in leprosy. It is characterized by the appearance of crops of brightly erythematous, tender, nodules or plaques with a tendency to bilateral symmetrical distribution. The extracutaneous manifestations include neuritis, iridocyclitis, orchitis and lymphadenopathy. Fever and other constitutional symptoms are usually associated with it. Vesicular or bullous lesions have also been reported rarely in severe type II reaction, mostly from Mexico and South America. From India there are few published reports about bullous reaction in leprosy and to the best of our knowledge, no case has so far been reported from Nepal. This type of lesion may give

Correspondence to: A. Rijal (e-mail: arpanaarjal@yahoo.co.uk)
diagnostic difficulty as it mimics many blistering disorders like pemphigus or bullous, necrotising, fixed drug eruption.

Case report

A 48-year-old male with lepromatous leprosy on WHO-MB-MDT developed high grade fever with rashes and was diagnosed as having the dapsone syndrome. He also developed bullous lesions on the trunk and extremities that ulcerated and healed, leaving behind atrophic scars. There was no history suggestive of exfoliative dermatitis, epistaxis, iridocyclitis, orchitis, neuritis, lymphadenopathy or hepatitis.

Cutaneous examination revealed multiple, hypopigmented, hypoaesthetic, bilaterally asymmetrical macules on the lower and upper extremities. Multiple atrophic scars were present on the lower limbs and buttocks. Madarosis was present. Bilateral thickened and non-tender ulnar nerve, radial cutaneous nerve and lateral popliteal nerve were noted. Glove and stocking type of anaesthesia with fixed flexion deformities of the small fingers of both hands were present. Atrophy of both thenar and hypothenar eminences was found.

All routine investigations were within normal limits except raised ESR (47 mm). His BI was 2-25. The patient was admitted to the ward and restarted WHO-MB-MDT. On the 4th day, he developed high-grade, intermittent fever with nausea, vomiting and loss of appetite, bone pain, myalgia, arthralgia and pedal oedema. Routine biochemical investigations showed leukocytosis. On day 10, a flaccid bulla was noted (Figure 1) and a slit skin smear from the lesion showed a BI of 3+. Histopathology of the lesion showed an intra-epidermal clef with diffuse polymorphonuclear infiltrate and a few foamy histiocytes in the dermis. Fite stain showed few AFB in the specimen. A diagnosis of lepromatous leprosy with bullous Type II reaction was made. The patient was started on prednisolone 60mg daily, clofazimine 100mg 8-hourly, but continued to get new lesions on the thigh, sole (Figure 2) and hand. From the 4th day onwards, his fever subsided and no new lesions appeared. He became asymptomatic after a week, the dose of prednisolone was tapered and he was discharged on 20mg daily. One month later, however, bullous lesions developed on the hands with boggy swelling of both tibias, pedal oedema, low back ache, high-grade fever and neuritis. High dose corticosteroid 60mg was given daily, but he continued to get new lesions and was therefore referred to a designated centre where thalidomide was available. After 2 days of thalidomide 100mg 8-hourly, no new lesions appeared. Thalidomide was gradually tapered down and the patient remained symptom free.

Discussion

Bullous lesions are not frequently seen in leprosy, though pustular, ulcerated and Erythema multiforme lesions have been reported, though the diagnosis was not confirmed by biopsy. Blistering disorders like pemphigus can occur suddenly without any preceding signs of inflammation. The oral mucosa is involved and when tangential pressure is applied to the periocular skin over a bony prominence there is separation of the epidermis from the dermis (Nikolsky’s sign). Bullous type reaction mimicking pemphigus in lepromatous leprosy without neuritis has been reported. Bullous ENL appearing de novo on the extremities

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may mimic bullous pemphigoid, which is always preceded by itching and weals. The diagnosis is again confirmed by histopathology.

Despite reservations from WHO on the use of thalidomide in leprosy,5 it has been described as the drug of choice for recurrent ENL.6 It is also a steroid sparing drug, with clinical response within 48–72 h. However, it has its shortcomings and has to be given under strict supervision to prevent misuse. It is not to be used in female patients planning a family

**Figure 1.** Vesicles in the right sole.

**Figure 2.** Bulla and erosions on left sole.
because of its teratogenic effect. Furthermore, in many leprosy endemic countries it is either banned, or not easily available. Physicians and leprosy health workers should be aware about this rare presentation of type II reaction because of its similarity with other blistering diseases and drug reaction.

References