Borderline Lepromatous Leprosy Presenting as a Single Cutaneous Plaque

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Sir,

The usual clinical features of lepromatous leprosy are multiple or symmetrical cutaneous lesions and symmetrical or widespread nervous involvement, which adversely affects the patient's quality of life. This disease may also sometimes develop without the patient's knowledge. In the early stage, a single cutaneous plaque might be the only obvious symptom. We describe here a case of a 23-year-old man with a single anaesthetic plaque on his right forearm, with no other sensory disturbances. The clinical presentation was consistent with tuberculoid leprosy, but a biopsy from the lesion revealed borderline lepromatous (BL) leprosy. The patient was started on multi-drug therapy for leprosy. Six months later the solitary anaesthetic cutaneous plaque became oedematous and erythematous, indicating a reversal reaction (type 1).

CASE REPORT

A 23-year-old Japanese Brazilian man, who had migrated to Japan 4 months earlier, noticed an irregular erythematous lesion on his right forearm in October 2006. The cutaneous lesion measured 10 cm in diameter and was slightly palpable and hypoaesthetic plaques (Fig. 1a). In particular, there were no thickened nerves in the vicinity of the forearm plaque. Results of routine laboratory examination were otherwise unremarkable. A test for anti-phenolic glycolipid-I in the serum was negative. Thus, tuberculoid leprosy was suspected clinically. However, a skin biopsy unexpectedly revealed changes consistent with BL-type leprosy: the dermis was expanded by a dense perivascular and periadnexal infiltrate of macrophages and lymphocytes, and the macrophages had granular and vacuolated cytoplasm (Fig. 2a). No collections of epithelioid histiocytes or multinucleate giant cells were observed. Fite-Faraco staining of sections revealed numerous acid-fast bacilli within the foam cells (Fig. 2b). Large numbers of acid-fast bacilli were also demonstrated by Ziehl-Neelsen staining, and anti-phenolic glycolipid-I staining was positive. No skin smears were prepared and the bacterial index[1] was not examined, because the patient's agreement could not be obtained. A DNA sequence from *Mycobacterium leprae* that encoded heat shock protein 70 was detected from the skin tissue by PCR (1). There were no mutations of the *rpo*β, *folP* or *gyrA* genes (2–4), which have been shown to be associated with resistance to rifampicin, dapsone, and fluoroquinolones, respectively. Based on these results and the histopathological findings, treatment was started with the combined standard multidrug therapy regimen and oral levofloxacin (300 mg daily). After 2 months' treatment, routine laboratory examination revealed mild anaemia, presumably caused by dapsone, and therefore dapsone was discontinued. Six months after the start of oral treatment, the patient developed a reversal reaction (type 1). The plaque on the left forearm became swollen and erythematous and the area of anaesthesia increased in size (Fig. 1b). The patient then returned to Brazil and has never returned to Japan.

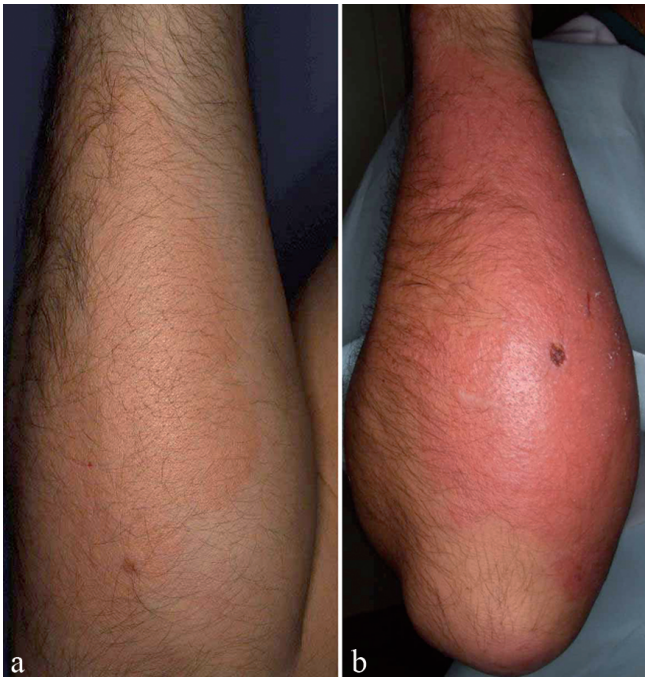

Fig. 1. (a) Clinical findings at first examination; a single, irregular erythematous lesion on the right forearm. (b) Reversal reaction; the lesion became swollen, erythematous and the area of anaesthesia increased.

DISCUSSION

BL-type leprosy is usually a generalized form of the disease with widespread skin infiltration, numerous macules, papules, plaques and/or nodules distributed symmetrically throughout the body (5). Clinically our case presented with a single lesion mimicking

[1]Bacterial index is simple and half quantity method of leprosy and it is obtained by surgical method, therefore patient's agreement is required.

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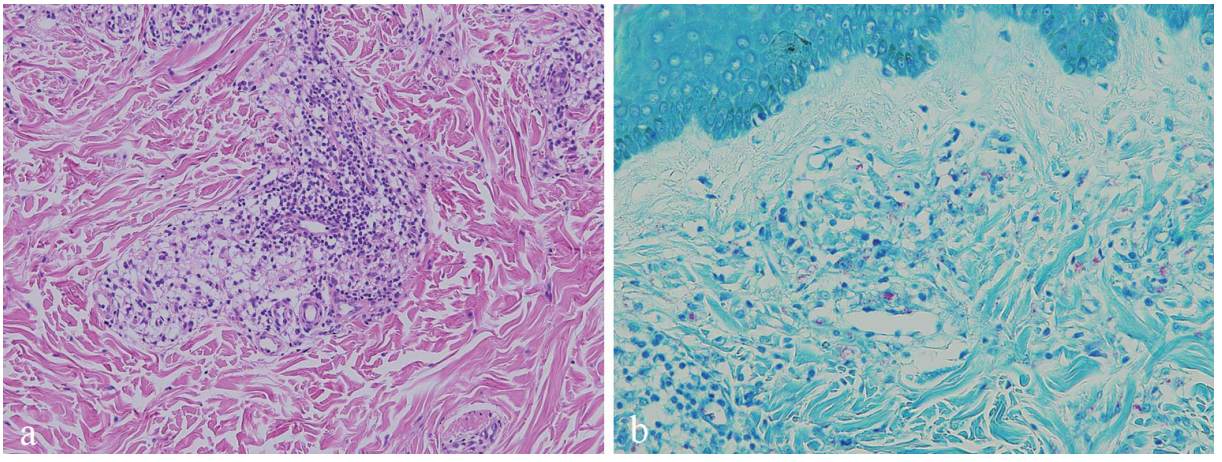

Fig. 2. Skin biopsy specimen; perivascular and perineural infiltration by numerous foamy cells in the dermis. (a) Haemotoxylin and eosin stain, original magnification ×80. (b) Fite-Faraco staining revealed numerous bacilli, original magnification ×120.

tuberculoid leprosy, although a biopsy from the lesion revealed BL-type leprosy, and the patient developed a reversal reaction, i.e. delayed hypersensitivity reaction resulting from immune recognition of the lepra bacilli within the dermal and truncal nerves (6), which due to multibacillary disease. The classification of leprosy might be based on histopathological findings and not clinical features (7). The presentation of multi-bacillary BL as a single cutaneous lesion or as localized lesions is rare and, to the best of our knowledge, there are four similar reports of cases in the literature that show a discrepancy between the histological findings and clinical features (5, 8–10). Patients with BL and lepromatous leprosy (LL)-type leprosy have been reported to develop type 1 or type 2 reactions (erythema nodosum) several months after the commencement of multidrug therapy. However, in 2 of 5 cases localized BL, patients presented only type 1 not type 2 reactions (9), and the reaction in other cases were not reported (5, 8, 10). These reports suggest that localized BL is still associated more with cellular immune reactions than those of BL or LL leprosy. The expression of multi-bacillary leprosy as a localized lepromatous lesion is a recognized but unusual presentation in leprosy. Possible explanations suggested for this presentation include direct exposure of the skin to *M. leprae* coupled with repeated trauma, low skin temperature and local immunological causes (11, 12), or the early-stage usual form of BL presents as a single lesion, which develops into multiple and symmetrical lesions over several months or years. There remains no convincing explanation as to how the highly bacillary forms of leprosy may be contained within a single lesion. The presentation of BL as a solitary plaque remains rare and reinforces the fact that certain aspects of the host cell-mediated response and pathophysiology of this disease are still not fully understood. Despite this, it is important that patients are classified correctly in the leprosy spectrum, so that they can be given the most appropriate treatment.

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