



Case report

Leprosy: A rare case of infectious peripheral neuropathy in the United States



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ABSTRACT

Peripheral neuropathy can be the initial presentation of leprosy. Diagnosis can be challenging unless skin manifestations are recognized. Skin biopsy and Fite staining are the keys to the diagnosis. It is important to treat coexisting Lepra reactions, peripheral neuropathy and side effects of the therapeutic agents. This is a complex clinical course of a patient with lepromatous leprosy.

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Introduction

The common infectious causes of peripheral neuropathy in New England area are HIV, EBV, CMV, HSV and Lyme disease [1]. We present a rare cause of peripheral neuropathy caused by an infection, leprosy.

Case

A 43-year-old Nepalese woman, who immigrated to the United States with her three children 2 years before the onset of symptoms, presented with burning pain in the soles of her feet for one month. Hemoglobin A1c, serum vitamin B12, and folate acid levels were normal. Electromyogram showed primary axonal peripheral distal neuropathy. Five months later, she started having erythematous nodular lesions over her face, the biopsy of which showed chronic inflammatory cells infiltrating into 30–40% of the dermis, and extending focally into the subcutaneous tissue (Fig. 1). Fite stains revealed a moderate number of clumped and intact acid fast bacilli (AFB) within histiocytes as well as within cutaneous nerve twigs (Fig. 2). The latter is pathognomonic for Hansen's disease. The bacillary index is 3–4+ and the presence of significant dermal edema is suggestive of type 1 reaction. Skin biopsy specimen was tested positive for *Mycobacterium leprae* and negative for *Mycobacterium lepromatosis* by polymerase chain

reaction (PCR). Physical examination shows multiple hypopigmented patches with hyperemic margins without much change in sensation over the trunk (Fig. 3). On her left leg, there was a non-tender, hypo-aesthetic violaceous erythematous geographic skin plaque, which later ulcerated (Fig. 4). It was clinically consistent with Lucio's phenomenon although no skin biopsy was done from that area.

This patient was diagnosed with Hansen's disease, mid-borderline (BB) with possible type-1 reaction and possible Lucio's phenomenon with peripheral neuropathy in lower extremities (Fig. 5). The patient was started on a multidrug regimen with dapsons 100 mg PO daily, clarithromycin 500 mg PO daily, moxifloxacin 400 mg PO once a month, prednisone 40 mg PO daily, methotrexate 10 mg PO weekly with folinic acid supplement (except on methotrexate Day) and vitamin D supplement 2000 IU PO daily [2–8].

At this writing, the patient is in the 12th month of therapy. Dapsone had to be switched to minocycline 100 mg PO daily in the 4th month of therapy due to the development of methemoglobinemia [9,10]. Methotrexate was stopped together with Dapsone. Prednisone has been tapered off towards the end of the 12th month of therapy. The patient developed a neuropathic ulcer over the left big toe during the 3rd month of therapy, which healed with wound care and off-loading. Lucio's phenomenon healed towards the end of 2nd month of therapy but left a scar. Hypopigmented skin patches disappeared totally within two months of therapy. Her youngest daughter (7-year-old) was treated with dapsons 25 mg PO daily and rifampin 150 mg PO monthly for a total of 12 months for the possible

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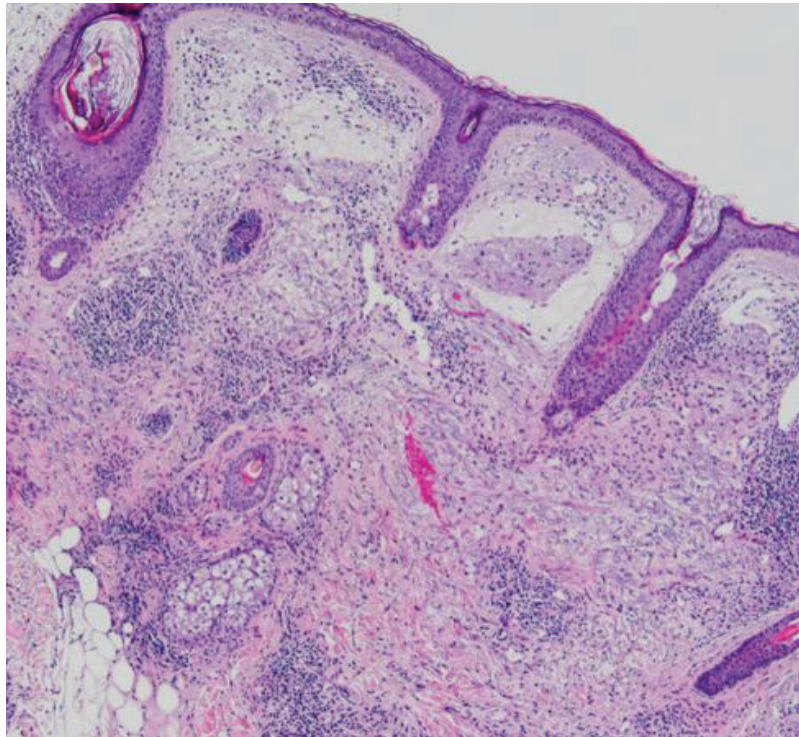


Fig. 1. Skin biopsy, H&E stain (low power): Chronic inflammatory cells (mononuclear) infiltrating dermis (yellow solid arrow) and focally into subcutaneous tissue.

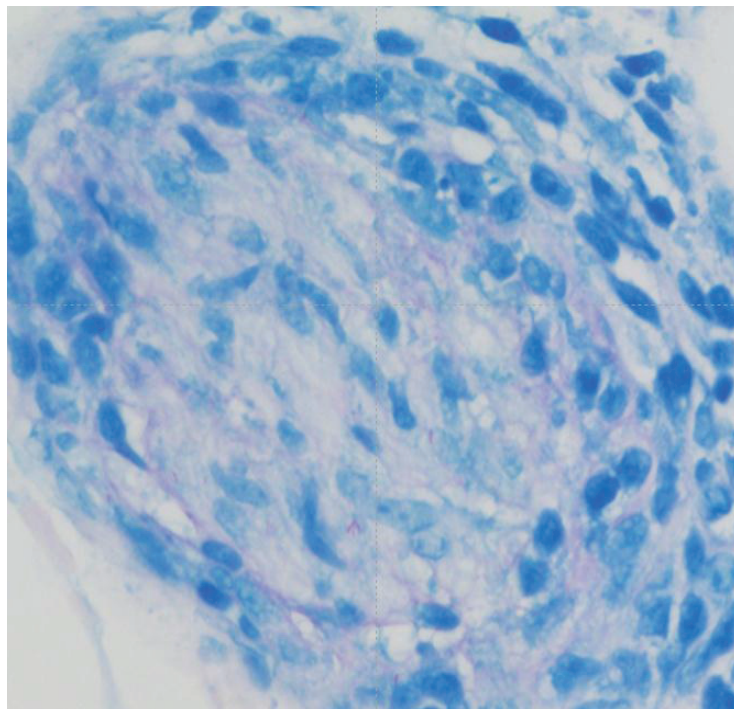


Fig. 2. Fite Stain, Dermal Nerve infiltrated by AFBs.

indeterminate form of leprosy [11]. Peripheral neuropathy in bilateral lower extremities persisted.

Discussion

Early recognition and prompt treatment are very important in Hansen's disease to be able to avoid sequelae such as

neuropathy and disfigurement [12]. It is essential to treat co-existing reactions both type 1 (reversal) and type 2 (Erythema Nodosum Leprosum ENL). The role of vitamin D supplementation is recognized recently in the treatment of leprosy [2–4]. Long tapering of steroid together with other immunomodulators such as methotrexate plays a major role in the treatment of lepra reactions [13–15]. Both reactions can present before,



Fig. 3. Hypopigmented skin patches with hyperemic margin over the back.



Fig. 4. Left leg: a non-tender, hypo-aesthetic violaceous erythematous geographic skin plaque, which later ulcerated: Possible Lucio's phenomenon.

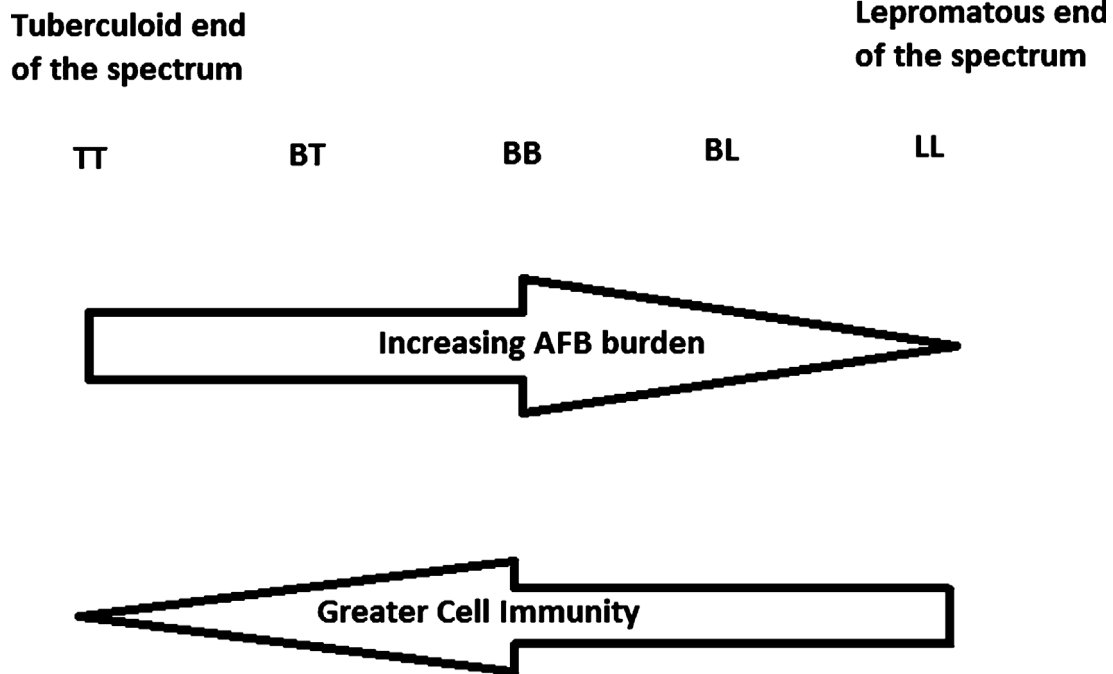


Fig. 5. Ridley-Jopling classification of leprosy, which reflects the spectrum of both clinical and pathological features. TT: tuberculoid; BT: borderline tuberculoid; BB: mid-borderline; BL: borderline lepromatous; LL: lepromatous; LL: lepromatous; AFB: acid fast bacilli.

during or after treatment with the multidrug regimen. Type 2 reaction also known as erythema nodosum leprosum (ENL) sometimes needs immunomodulatory drugs for many years depending on the clinical spectrum of Hansen's disease [13–15]. Screening of household contacts is recommended once a patient is diagnosed with Hansen's disease. Prevention and treatment of neuropathic ulcers are part of the treatment strategy for Leprosy. Multi-disciplinary involvement such as podiatry, wound care, neurology, dermatology, infectious disease, and psychosocial support is the key to treatment success.

Consent

Written informed consent was obtained from the patient for the publication of the case. A copy of the consent is available for review on request.

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None.

CRedit authorship contribution statement

Ye Min Oo: Conceptualization, Formal analysis, Writing - original draft, Writing - review & editing. **Armando Paez:** Formal analysis, Writing - review & editing, Supervision. **Richard Brown:** Formal analysis, Writing - review & editing, Supervision.

Declaration of Competing Interest

None.

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