Atypical Presentation of Borderline Tuberculoid (BT) Leprosy in an Immunocompetent Adult – A Case Report

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Abstract

Leprosy is a chronic and progressive granulomatous disease that primarily affects the skin and peripheral nerves and is caused by the bacterium Mycobacterium leprae. Despite its long history with humans, its varied presentation continues to baffle clinicians. However, clinicians may miss many cases due to decreased clinical suspicion of leprosy and atypical case presentations. We present an unusual case of borderline tuberculoid leprosy, which presented as chronic skin ulcers with no associated anaesthetic skin patches, peripheral nerve thickness and neuropathy, mimicking cutaneous leishmaniasis. This case report highlights atypical presentations of borderline tuberculoid leprosy that clinicians may miss without a high index of suspicion and or skin biopsy.

A 54-year-old male trader presented with a 2-year history of a chronic ulcer on the dorsum of his right hand and an 8-month history of a similar ulcer on his nose. Both lesions began as painless, non-pruritic swellings. Examination revealed a 12 cm x 11 cm ulcer on the right hand and a 7 cm x 5 cm ulcer on the nose. No loss of sensation or nerve enlargement was detected. Routine investigations, including complete blood count, renal function test and HIV screening, were negative. A slit skin smear revealed acid-fast bacilli, and a skin biopsy suggested BT leprosy. There was a remarkable improvement in symptoms after multibacillary anti-leprosy treatment.

Ulceration is an unusual presentation of borderline tuberculoid leprosy. This case highlights the importance of maintaining a high index of suspicion and the role of skin biopsy in the prompt diagnosis and treatment of similar cases.

Keywords: Atypical presentation, Borderline tuberculoid, Leprosy, Atrophic ulcers

Présentation atypique de la lèpre tuberculoïde borderline (TB) chez un adulte immunocompétent à propos d'un cas

La lèpre est une maladie granulomateuse chronique et évolutive qui affecte principalement la peau et les nerfs périphériques, causée par la bactérie Mycobacterium leprae. Malgré sa longue histoire chez l'homme, sa présentation variée continue de dérouter les cliniciens. Cependant, de nombreux cas peuvent être manqués par les cliniciens en raison d'une suspicion clinique réduite et de présentations atypiques. Nous présentons un cas inhabituel de lèpre tuberculoïde borderline, qui se présentait sous forme d'ulcères cutanés chroniques sans plaques cutanées anesthésiantes associées, d'épaississement des nerfs périphériques et de neuropathie, simulant une leishmaniose cutanée. Ce cas clinique met en évidence les présentations atypiques de la lèpre tuberculoïde borderline que les cliniciens peuvent manquer sans une forte suspicion et/ou une biopsie cutanée.

Il s'agit d'un commerçant de 54 ans qui a consulté pour un ulcère chronique sur le dos de la main droite évoluant depuis deux ans et un autre sur le nez depuis huit mois. Les deux lésions ont débuté par des gonflements indolores et non prurigineux. L'examen a révélé un ulcère de 12 cm x 11 cm sur la main droite et un autre de 7 cm x 5 cm sur le nez. Aucune perte de sensibilité ni hypertrophie nerveuse n'a été détectée. Les

examens de routine, dont une numération formule sanguine, un bilan rénal et un dépistage du VIH, se sont révélés négatifs. Un frottis cutané a révélé la présence de bacilles acido-résistants, et une biopsie cutanée a suggéré une lèpre BT. Une amélioration remarquable des symptômes a été observée après un traitement antilépreux multibacillaire.

L'ulcération est une manifestation inhabituelle de la lèpre tuberculoïde borderline. Ce cas souligne l'importance d'une forte suspicion et le rôle de la biopsie cutanée dans le diagnostic et le traitement rapides de cas similaires.

Mots-clés: Présentation atypique, Tuberculoïde borderline, Lèpre, Ulcères atrophiques

Background

Leprosy, also known as Hansen's disease, is a chronic granulomatous infection caused by *Mycobacterium leprae* and mainly transmitted from one person to another by aerosol droplets through the nasal mucosa.[1] It primarily affects the peripheral nerves and skin.[1] According to the World Health Organization (WHO), individuals having one of the following three features are considered to have leprosy: (i) definite loss of sensation in a pale (hypopigmented) or reddish skin patch; (ii) a thickened or enlarged peripheral nerve with loss of sensation; and (iii) the presence of acid-fast bacilli in a slit-skin smear.[1] This disease is classified according to the Ridley-Jopling classification into five main variants based on clinical and histopathological features, including tuberculoid (TT), borderline tuberculoid (BT), mid-borderline (BB), borderline lepromatous (BL), and lepromatous types.[2] The Borderline tuberculoid (BT) leprosy is clinically characterized by symmetrically distributed hypopigmented patches varying in size from 2 to 20 cm and thickened peripheral nerves.[3]

Other possible clinical presentations of BT include acute painful neuritis, ulcers, loss of neurological function, thickening of nerves, abscesses, high fever, and sudden urticaria of leprotic skin lesions resulting from increased inflammation of preexisting lesions.[4]

Ulcers are not a typical early clinical presentation in leprosy patients, except during reactional states, Lucio's phenomenon (LP), or secondary to neuropathies.[5] A non-healing ulcer was reported in a 70-year-old lady with borderline tuberculoid Leprosy from India, which healed following anti-leprosy medications.[6]

We report a case of BT leprosy presenting with multiple ulcers on the face and the hand, which is an unusual presentation of BT leprosy. This case is one of the rare instances where ulcers are the primary clinical feature of BT leprosy.

Case Report

A 54-year-old trader presented with a two-year history of multiple ulcers on the dorsum of the right hand and the nose. The patient first noticed the ulcer on the dorsum of his right hand, which began as a small, raised papule that gradually increased in size and subsequently developed into an ulcer. A similar lesion on the nose had developed 8 months prior to presentation. The lesions were not associated with pain, itching, or a burning sensation. There was no loss of sensation, and no skin patches were observed. There is no history of similar lesions in any family member or community. There is no history of similar lesions; the patient has never been treated for leprosy. There was no history of contact with a known case of leprosy, and he does not have any background medical condition.

On clinical examination, there was an ulcer on the dorsum of the right hand extending from the wrist to the knuckle, with sloping edges measuring about 12cm by 11cm [Figure 1a] and another ulcer on the nose extending from the proximal nasal bridge to the tip of the nose also measuring 7cm by 5cm [Figure 1a]. No anaesthetic skin patch or peripheral nerve enlargement was observed.

Histopathological examination of the skin ulcer showed an ulcerated pigmented stratified squamous epithelium overlying a collagenous dermis densely infiltrated by diffuse granulomata composed of macrophage, giant cells and intense lymphocytic infiltrates surrounding adnexal and nerve bundles

(perineural and periadnexal). Special stains for acid-fast bacilli (AFB), using the Ziehl–Neelsen (ZN) and Fite-Faraco methods, were unhelpful. [Figure 2a]. Slit skin smears of the ear lobe revealed characteristic acid-fast bacilli (AFB) in large numbers [Figure 2b]. The findings supported the microbiological diagnosis of leprosy. The patient was considered as a case of Borderline Tuberculoid Leprosy and started on multidrug therapy (MDT).

The Skin ulcers improved rapidly (within 4 weeks) after the commencement of multidrug therapy for paucibacillary Leprosy (MDT-PB) using dapsone and rifampicin, according to WHO recommendations. This resulted in the complete healing of the nasal ulcer and the formation of granulation tissue on the hand ulcer. (Figures 1b and 1c). The patient is being considered for a skin graft of the hand ulcer.

Discussion

Patients with BT leprosy rarely present with skin ulcers except during reaction states, Lucio's phenomenon, or as a complication of peripheral neuropathy, where the ulcer is commonly seen on the plantar surface of the foot or the upper extremities.[5] In this index case, the ulcer was observed on the face and the upper extremity and rapid healing was noticed after the commencement of MDT-MB. There was also no associated feature to suggest peripheral neuropathy or nerve enlargement, which are unusual presentations of BT. This ulcer is different from neuropathic ulcers seen as complications of peripheral neuropathy in leprosy, with very slow healing of the ulcers due to the vascular and neural disabilities that underlie these ulcers, requiring specific therapeutic interventions in addition to MDT-MB.[5,7] Non-neuropathic ulceration in leprosy is not frequently found. Thus, there is a need for a high index of suspicion and detailed workup to arrive at the diagnosis and initiate appropriate therapy.

In this case report, we have demonstrated a cause of non-trophic ulceration in leprosy, specifically spontaneous ulceration in BT, which presented as superficial ulcers over pre-existing erythematous and edematous plaques around the nose, a rare manifestation of ulceration in leprosy. The early manifestation of leprosy as non-trophic skin ulceration may pose a diagnostic delay or even a misdiagnosis, as the index case has been with the ulcer for 2 years while using several antibiotics with little or no improvement, even at presentation to our clinic, we never thought of leprosy initially due to absence of other cardinal features to suggest it.

Some differential diagnoses of BT leprosy that should be considered here include lupus vulgaris (LV), sarcoidosis, granuloma faciale, lymphocytoma cutis, and mycosis fungoides.[8] This was one of the reasons why slit skin smears and skin biopsy were done to confirm the diagnosis of borderline tuberculoid leprosy because of the atypical presentation in this index patient.

With histopathological examination, leprosy presents as granulomatous inflammation that may mimic other granulomatous diseases. Special stains such as Fite–Faraco stain have an added advantage in demonstrating *M. leprae* in histopathological examination but may show negative results in tuberculoid leprosy as shown in this atypical case of BT where the skin lesions show histopathological features in keeping with BT, though the slit skin smear has demonstrated AFB.[9]

Conclusion

This case report has highlighted the importance of recognising non-trophic ulcers as a possible atypical skin manifestation of borderline leprosy without clinical manifestation of neuropathy. Therefore, clinicians are encouraged to consider leprosy as a differential diagnosis of all ulcers that are not healing, especially in the tropics where leprosy is endemic. There is also a need for a clinico-histopathological correlation of all chronic non-trophic ulcers, allowing for early diagnosis and prompt treatment of leprosy to avoid disabilities.

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Conflicting Interest (If present, give more details):
None

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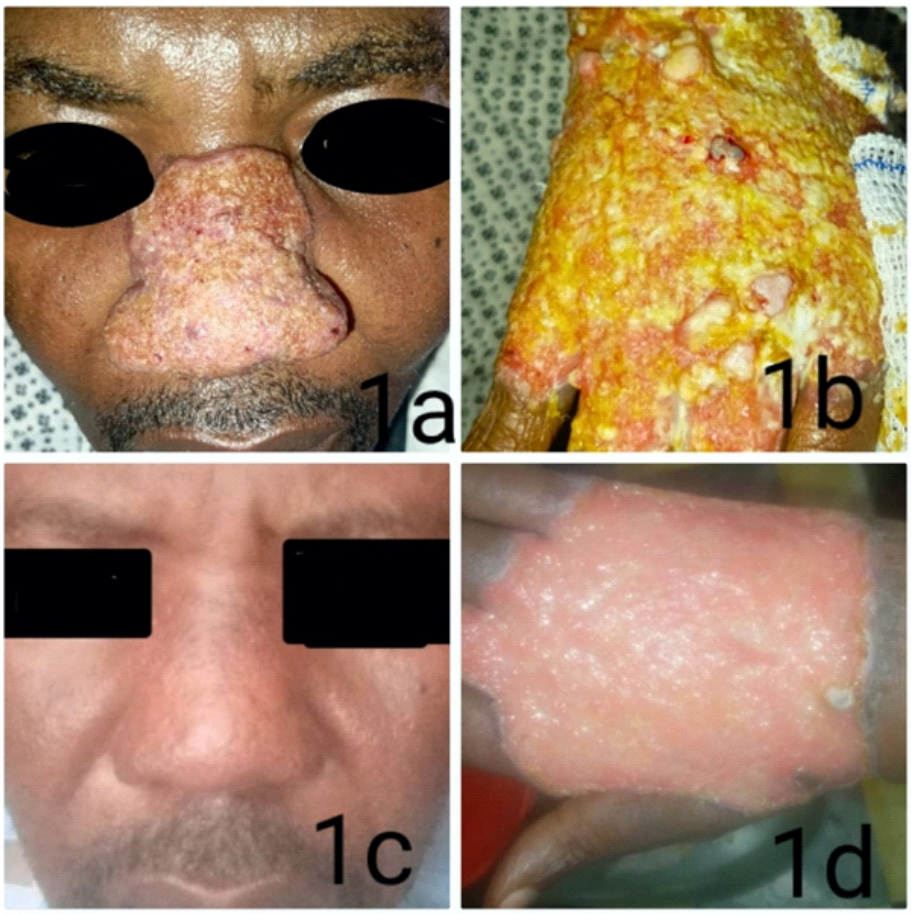

Figure 1a/1b (nose and hand ulcers before treatment),
Figure 1c/1d (healed nose ulcer after treatment and hand ulcer forming granulation tissues)

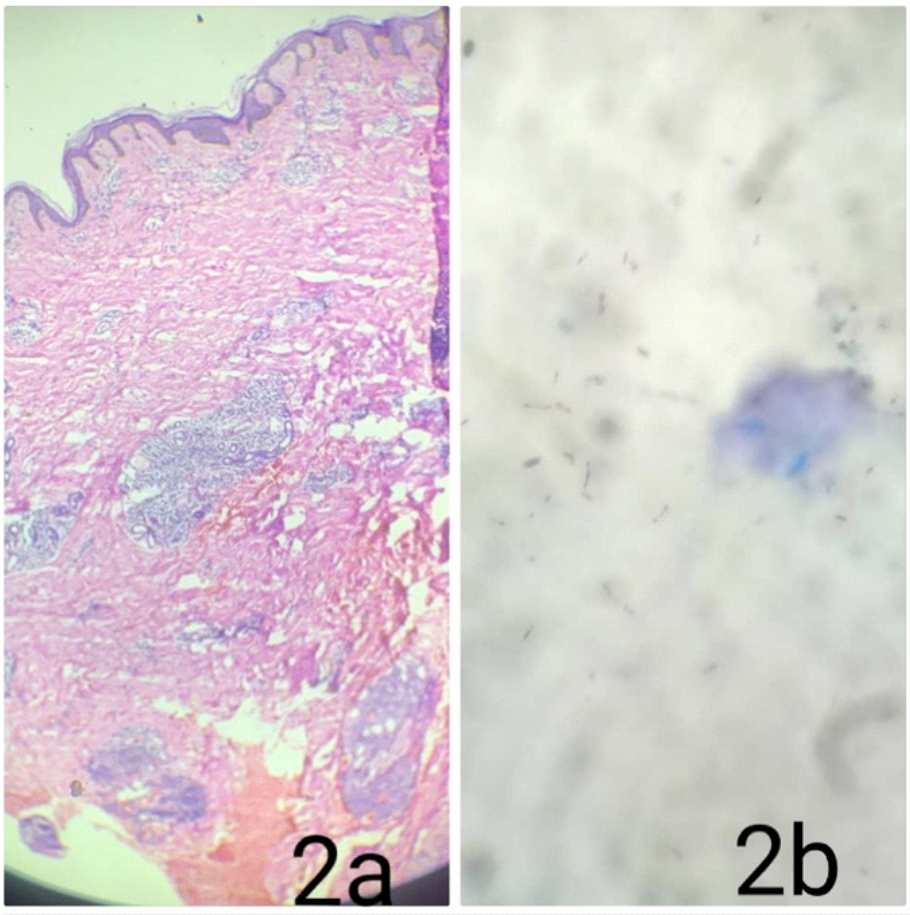

Figure 2a: shows the histology of ulcer x40,
Figure 2b: shows multiple Acid-fast bacilli from slit skin smear x40