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CASE REPORTS ON LEPROSY – A SHARED PRACTICE

Ulcerative type 1 lepra reaction in borderline-tuberculoid leprosy: a rare presentation

Reação tipo 1 ulcerativa em hanseníase dimorfo-tuberculóide: uma apresentação rara

Reacción tipo 1 ulcerativa en la lepra dimorfo-tuberculoide: una presentación rara

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ABSTRACT

Introduction: leprosy is a chronic infectious disease caused by *Mycobacterium leprae*, characterized by a wide spectrum of clinical presentations. In India, borderline-tuberculoid leprosy is the most common form encountered in clinical practice. Type 1 lepra reaction in borderline-tuberculoid leprosy usually presents as the development of erythema and/or edema in pre-existing skin lesions. Ulceration of skin lesions in type 1 lepra reaction is uncommon and occurs in severe reactions. **Objective:** to report an unusual presentation of borderline-tuberculoid leprosy with ulcerative type 1 lepra reaction in an immunocompetent patient. **Case description:** we present the case of a 65-year-old man with chief complaints of ulcerated plaque over his left thigh. He also had other skin lesions suggestive of borderline-tuberculoid leprosy over his trunk and limbs, as well as

enlarged, mildly tender left ulnar and lateral popliteal nerves. A slit skin smear was negative, while a skin biopsy supported the diagnosis of borderline-tuberculoid leprosy. The patient responded to multibacillary multidrug therapy according recommended by World Health Organization and tapering doses of prednisolone, with complete healing of the ulceration at six weeks follow-up. **Discussion:** type 1 lepra reaction associated with borderline-tuberculoid leprosy usually presents with increased erythema and edema in pre-existing skin lesions. Ulceration in such skin lesions is not commonly seen except in cases with severe type 1 leprosy reactions. Administration of oral corticosteroids along with multibacillary multidrug therapy is the key to managing ulcerative type 1 lepra reaction. The ulceration heals rapidly with tapering doses of oral corticosteroids, limiting the duration of morbidity. **Final consideration:** the case emphasizes the need for dermatologists and leprologists to be aware of atypical presentations of leprosy reactions, ensuring timely diagnosis and effective management to achieve optimal patient outcomes.

Keywords: *Leprosy. Type 1 Reaction. Lazarine Leprosy. Borderline-Tuberculoid Leprosy.*

RESUMO

Introdução: a hanseníase é uma doença infecciosa crônica causada pelo *Mycobacterium leprae*, caracterizada por um amplo espetro de manifestações clínicas. Na Índia, a hanseníase dimorfa-tuberculoide é a forma mais comum encontrada na prática clínica. A reação hansênica tipo 1, nos pacientes dimorfo-tuberculoide, manifesta-se normalmente com a presença de eritema e/ou edema em lesões cutâneas pré-existentes. A ulceração das lesões cutâneas na reação tipo 1 é pouco frequente e ocorre em reações graves. **Objetivo:** relatar uma apresentação atípica de hanseníase dimorfa-tuberculoide com reação tipo 1 ulcerada em paciente imunocompetente. **Descrição do caso:** apresentamos o caso de um homem de 65 anos com queixa principal de placa ulcerada na coxa esquerda. Apresentava também, outras lesões cutâneas sugestivas de hanseníase dimorfa-tuberculoide no tronco e nos membros, bem como os nervos ulnar e poplíteo lateral esquerdos espessados e levemente doloridos. A baciloscopia foi negativa, enquanto a biópsia de pele confirmou o diagnóstico de hanseníase dimorfa-tuberculoide. O paciente respondeu à terapêutica com poliquimioterapia multibacilar, recomendada pela Organização Mundial de Saúde, e a doses decrescentes de prednisolona, com cicatrização completa da ulceração após 6 semanas de seguimento. **Discussão:** a reação hansênica tipo 1 associada a hanseníase dimorfa-tuberculoide apresenta-se geralmente com aumento de eritema e edema em lesões cutâneas pré-existentes. A ulceração dessas lesões não é comum, exceto nos casos graves. A administração de corticosteróides orais associada a poliquimioterapia multibacilar é a chave para

o tratamento da reação hansênica tipo 1 ulcerada. A cicatrização da ulceração ocorre rapidamente com doses decrescentes de corticosteróides orais, limitando a duração da morbilidade. **Consideração final:** o caso enfatiza a necessidade de os dermatologistas e hansenologistas estarem atentos às apresentações atípicas das reações hansênicas, assegurando um diagnóstico oportuno e um manejo eficaz para alcançar os melhores resultados na evolução dos pacientes.

Palavras-chave: *Hanseníase. Reação Tipo 1. Hanseníase Lazarina. Hanseníase Dimorfa-tuberculóide.*

RESUMEN

Introducción: la lepra es una enfermedad infecciosa crónica causada por *Mycobacterium leprae*, caracterizada por un amplio espectro de manifestaciones clínicas. En la India, la lepra dimorfo-tuberculoide es la forma más frecuente en la práctica clínica. La reacción leprosa de tipo 1 en pacientes con lepra dimorfo-tuberculoide suele manifestarse con la presencia de eritema y/o edema en lesiones cutáneas preexistentes. La ulceración de las lesiones cutáneas en las reacciones de tipo 1 es rara y se produce en las reacciones graves. **Objetivo:** Comunicar una presentación atípica de lepra dimorfo-tuberculoide con reacción ulcerada de tipo 1 en un paciente inmunocompetente. **Descripción del caso:** presentamos el caso de un hombre de 65 años con la queja principal de una placa ulcerada en el muslo izquierdo. También presentaba otras lesiones cutáneas sugestivas de lepra dimorfo-tuberculoide en el tronco y las extremidades, así como nervios cubital y poplíteo lateral izquierdos engrosados y ligeramente dolorosos. La baciloscopia fue negativa, mientras que la biopsia cutánea confirmó el diagnóstico de lepra dimorfo-tuberculoide. El paciente respondió a la terapia con politerapia multibacilar, como recomienda la Organización Mundial de la Salud, y dosis decrecientes de prednisolona, con curación completa de la ulceración tras 6 semanas de seguimiento. **Discusión:** la reacción leprosa de tipo 1 asociada a la lepra dimorfo-tuberculoide suele presentarse con aumento del eritema y edema en lesiones cutáneas preexistentes. La ulceración de estas lesiones es infrecuente, excepto en los casos graves. La administración de corticoesteroides orales asociada a una politerapia multibacilar es la clave del tratamiento de la reacción ulcerada de tipo 1 de la lepra. La ulceración cicatriza rápidamente con dosis decrecientes de corticoides orales, limitando la duración de la morbilidad. **Consideración final:** el caso subraya la necesidad de que los dermatólogos y especialistas en lepra estén atentos a las presentaciones atípicas de las reacciones leprosas, asegurando un diagnóstico a tiempo y un tratamiento eficaz para conseguir los mejores resultados en la evolución de los pacientes.

Palabras clave: *Lepra. Reacción Leprosa. Reacción de Tipo 1. Lepra Lazarina. Lepra Dimorfo-tuberculoide.*

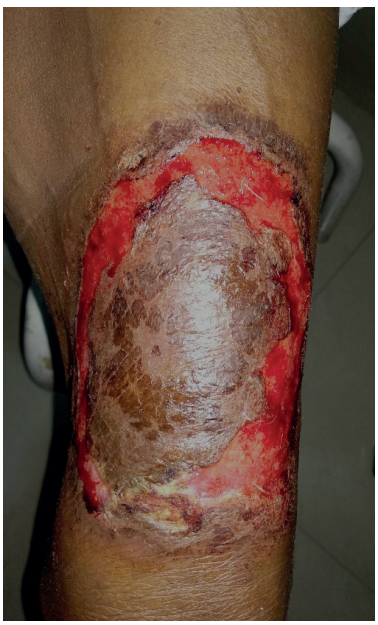
INTRODUCTION

The chronic course of leprosy is often interrupted by episodes of immunologically mediated type 1 and type 2 leprosy reactions[1]. Type 1 leprosy reaction is commonly seen in the borderline group, borderline-tuberculoid, and mid-borderline leprosy, while type 2 leprosy reaction or erythema nodosum leprosum is seen in lepromatous leprosy. Type 1 reaction mainly involves the skin and nerves, whereas type 2 reaction is associated with systemic features in addition to skin and nerve involvement. Ulcerative and necrotic lesions are well described in type 2 reaction or erythema nodosum leprosum (ENL) but not in type 1 reaction[2]. The usual presentation in type 1 leprosy reaction is an increase in erythema and/or edema in already existing lesions. A severe type 1 reaction may develop ulcerative lesions, an extremely rare occurrence.

CASE PRESENTATION

A 65-year-old man presented to the leprosy clinic with chief complaints of an erythematous plaque over the left thigh for three months with the development of increased erythema and ulceration for ten days. There was minimal pain and burning sensation associated with the ulcerated lesion. There was no history of fever, arthralgias, nerve pain, or motor weakness. General physical examination was within normal limits. On cutaneous examination, there were a few erythematous plaques of varying sizes over the trunk and limbs. The largest plaque was present over the anterior aspect of the left thigh, measuring approximately 15 × 8 cm (Figure 1).

Figure 1 – Erythematous ulcerated plaque over the thigh.



Source: Created by the author.

The periphery of the plaque was ulcerated with oozing blood in places but with a clean base. Sensory examination revealed partial loss of temperature and pain sensations at the central part of the plaque. Left ulnar and left lateral popliteal nerves were found to be enlarged and mildly tender. Sensations over the hands and feet were intact, and motor examination was normal. Based on the clinical findings, a diagnosis of borderline-tuberculoid leprosy with ulcerative type 1 leprosy reaction was made. Slit skin smear was negative, and histopathology of the skin lesion revealed tuberculoid granuloma consistent with the diagnosis of BT leprosy (Figure 2 & Figure 3).

Figure 2 – Photomicrographs showing nerve bundles and non-caseating granuloma (H&E, ×10).

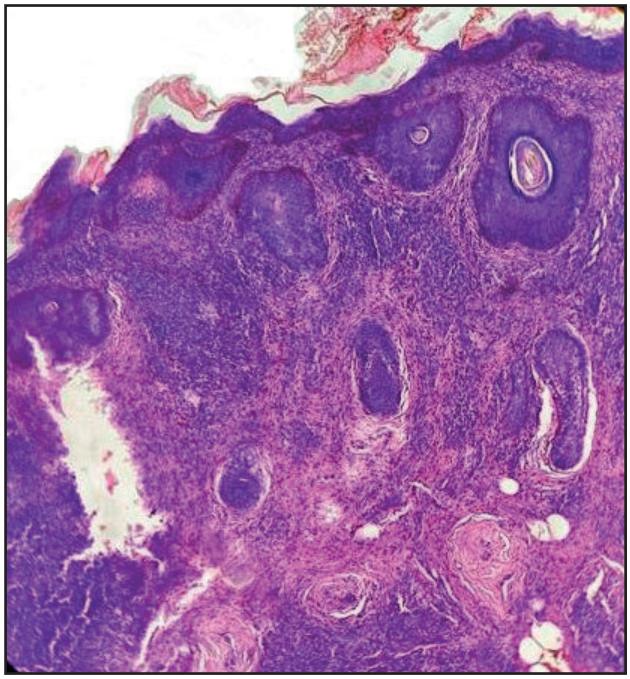
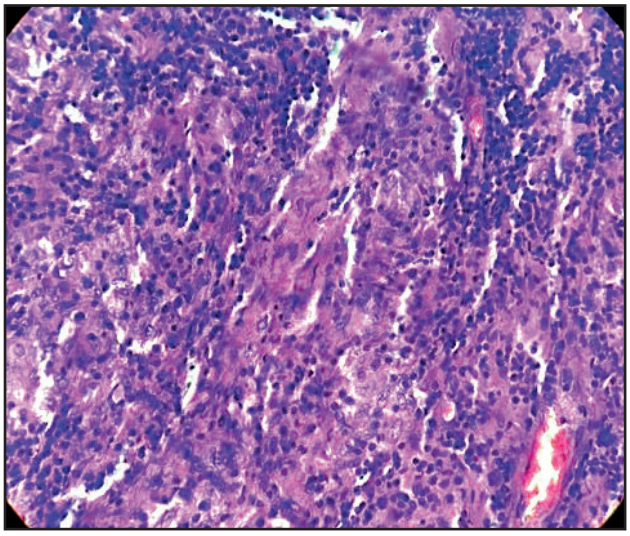


Figure 3 – Photomicrographs showing epitheloid cell granuloma with dense inflammatory mononuclear cell infiltrate (H&E, ×100).



All blood investigations were within normal limits, and HIV serology was negative. The patient was started on WHO multibacillary multidrug therapy (MDT), comprising monthly supervised doses of rifampicin 600 mg and clofazimine 300 mg, daily administered dapsone 100 mg and clofazimine 50 mg, and oral prednisolone at a dose of 1 mg/kg/day tapered over 12 weeks. The ulcer started healing within two weeks, and complete healing was seen at six weeks with post-inflammatory hypopigmentation and slight atrophy (Figure 4 & 5).

Figure 4 – At a two-week follow-up, the ulcer started healing.

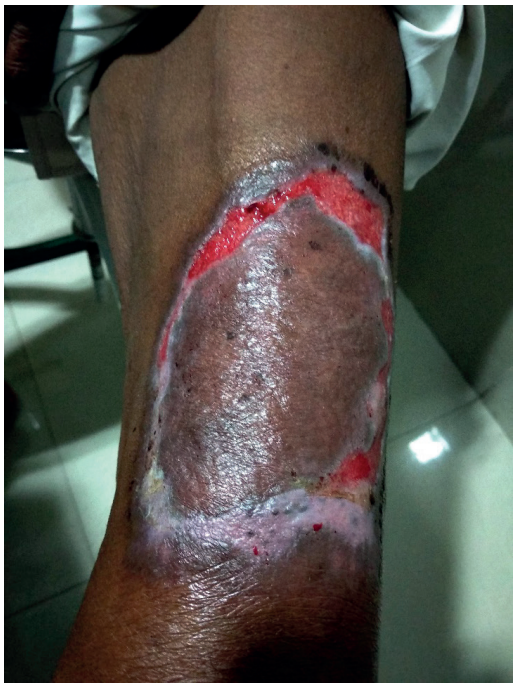
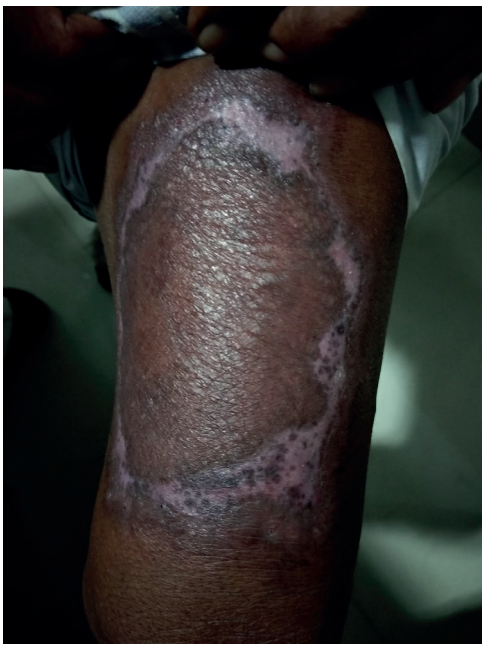


Figure 5 – At six weeks' follow-up, complete healing was achieved with post-inflammatory hypopigmentation and slight atrophy.



DISCUSSION

Leprosy is a chronic infectious granulomatous disease caused by *Mycobacterium leprae*[3]. It is one of the WHO's neglected tropical diseases with a variety of clinical manifestations and long-term complications and disabilities[4]. Depending upon the host immunity, the clinical picture presents along a spectrum with a tuberculoid pole at one end and a lepromatous pole at the other end. The various types of leprosy with distinct clinical presentation, bacteriology, immunological status, and histopathological findings include tuberculoid (TT), borderline-tuberculoid (BT), mid-borderline (BB), borderline-lepromatous (BL) and lepromatous leprosy (LL)[5].

The most common type of leprosy encountered in India is borderline-tuberculoid[4]. It usually presents as hypopigmented, hypoaesthetic macules or plaques, which can have an annular configuration in many cases. Satellite lesions and pseudopodia are also described with BT leprosy. The borderline leprosy group is considered unstable and is more prone to develop leprosy reactions. Type 1 leprosy presents as increased erythema and/or edema of pre-existing skin lesions with or without neuritis. Type 2 reaction presents as evanescent tender nodules with systemic fever symptoms and arthralgias. Although commonly seen in type 2 reactions (ENL necroticans), ulcerative lesions are unusual in type 1 leprosy reactions. The skin lesions in type 1 leprosy reaction rarely undergo ulceration, which is believed to be due to an exaggerated immune response[2]. This has

been termed as 'Lazarine leprosy' by some authors[1,6,7]. Oral corticosteroids are the mainstay of management in type 1 leprosy reaction. The ulcerative lesion heals completely with no or minimal scarring.

Our case describes an unusual type 1 leprosy reaction presentation with excellent response to multidrug therapy and oral corticosteroids. Such presentation can also occur in BB and BL types, sometimes in association with HIV infection[8]. Treatment with steroids provides an excellent response with complete healing of ulcers. Healing of ulcers can leave behind atrophic scarring if the ulcers are deep. Mushtaq[9] reported a case of ulcerative type 1 reaction healing with morpheaform scarring, unlike our case, which showed post-inflammatory hypopigmentation with minimal scarring.

CONCLUSION

This case is reported here due to its unusual presentation. Dermatologists and leprologists should be aware of such atypical presentations of leprosy reactions for prompt diagnosis and timely treatment.

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REFERENCES

1. Sunandini PA, Prasad PG, Chalam KV, Padmasiri Y. Type 1 lepra reaction with ulceration (Lazarine leprosy): two interesting case reports. IOSR J Dent Med Sci. 2015 [cited 2023 Jul 12];14(1):22-5. Available from: https://www.iosrjournals.org/iosr-jdms/papers/Vol14-issue1/Version-4/F014142225.pdf.

2. Hegde P, Jaiswal D, Shetty VM, Pai K, Rao R. Cutaneous ulceration as a presenting feature of type 1 lepra reaction: a case report. Trop Doct. 2022;52(2):354-6. doi: https://doi.org/10.1177/00494755211068661.

3. Jopling WH, McDougall AC. Handbook of leprosy. 5th ed. New Delhi: CBS Publishers; 1996. p. 1-7.

4. Mushtaq S, Dogra N, Dogra D, Faizi N. Trends and patterns of leprosy over a decade in a tertiary care hospital in Northern India: A retrospective analysis. Indian J Dermatol Venereol Leprol. 2020;86(2):141-9.

5. Kumar B, Dogra S. Case definition and clinical types of leprosy. In: Kumar B, Kar HK. IAL Textbook of Leprosy. 2nd ed. New Delhi: Jaypee; 2017. p. 238-50.

6. Wankhade V, Shah V, Singh RP, Bhat D. Lazarine leprosy: a unique phenomenon of leprosy. Int J Mycobacteriol. 2020;9(3):329-31. doi: https://doi.org/10.4103/ijmy.ijmy_104_20.

7. Bhat R, Pinto M, Dandakeri S, Kambil S. Ulcerating type 1 lepra reaction mimicking Lazarine leprosy: an unusual presentation of immune reconstitution inflammatory syndrome in an HIV-infected patient. Int J STD AIDS. 2013;24(12):992-4. doi: https://doi.org/10.1177/0956462413490144.

8. Mouchard A, Blaizot R, Graille J, Couppie P, Bertin C. Leprosy as immune reconstitution inflammatory syndrome in patients living with HIV: description of French Guiana's cases over 20 years and systematic review of the literature. PLoS Negl Trop Dis. 2022;16(3):e0010239. doi: https://doi.org/10.1371/journal.pntd.0010239.

9. Mushtaq S. Ulcerated cutaneous lesions in type 1 lepra reaction healing with morpheaform scarring: an unusual presentation. Int J Dermatol. 2023;62(6):e350-1. doi: https://doi.org/10.1111/ijd.16586.