doi: http://dx.doi.org/10.11606/issn.1679-9836.v104i5e-231502 Rev Med (São Paulo). 2025 Sept-Oct;104(5):e-231502.

Case Report

Linear aspect of borderline leprosy in a female patient: an atypical manifestation of a skin lesion

Aspecto linear da hanseníase borderline em paciente do sexo feminino: uma apresentação atípica de lesão no tegumento

Nivin Mazen Said[1], Airton Kenji Motizuki[2], Nirlando Igor Fróes Miranda[3], Marília Brasil Xavier[4]

Said NM, Motizuki AK, Miranda NIF, Xavier MB. Linear aspect of borderline leprosy in a female patient: an atypical manifestation of a skin lesion / *Aspecto linear da hanseníase borderline em paciente do sexo feminino: uma apresentação atípica de lesão no tegumento.* Rev Med (São Paulo). 2025 Sept-Oct;104(5):e-231502.

ABSTRACT: Leprosy is a chronic infection influenced by the immune response of each host, and its inflammatory immunopathology can present both neural and dermatological manifestations. This case involves a 17-year-old female patient, phototype IV, diagnosed with leprosy, who developed a hypochromic, erythematous, infiltrated linear plaque along the ulnar pathway, accompanied by claw hand deformity in the right upper limb. Additionally, complementary examinations, including histopathological analysis, confirmed the diagnosis of borderline leprosy. During the second month of clinical follow-up, the patient's β-HCG test returned positive, indicating pregnancy. Literature suggests that the heterogeneous cutaneous presentation observed in DD leprosy reflects an intensely variable immune response, with lesions oscillating between the multibacillary and paucibacillary poles of the disease spectrum. This leprous lesion exhibits a unique aspect not documented in the literature, highlighting the need for further investigation into these diverse manifestations in order to prevent misdiagnosis and facilitate early treatment.

KEY WORDS: Leprosy; Linear lesion; Borderline; Ulnar path.

RESUMO: A hanseníase é uma infecção crônica dependente da reação imunológica de cada hospedeiro e sua imunopatologia de caráter inflamatório caracteriza-se pelo acometimento neural e dermatológico bem como sequelas motoras potencialmente irreversíveis. O caso em questão é de uma paciente de 17 anos, fototipo IV, diagnosticada com hanseníase que evoluiu com uma placa hipocrômica e eritematosa, infiltrada em formato linear de seguimento do trajeto ulnar juntamente com a mão em garra no membro superior direito. Foram realizados exames complementares como o anatopatológico que confirmou o diagnóstico de hanseníase dimorfa dimorfa (DD) e durante o segundo mês de seguimento clínico, o exame beta HCG da paciente teve resultado positivo para gravidez. Estudos indicam que a variada apresentação cutânea da hanseníase, especialmente na forma DD, deve-se à resposta imunológica intensamente variável dessa forma clínica, que apresenta uma oscilação entre os dois polos da doença, multibacilar e paucibacilar. Esta lesão hansênica apresenta um aspecto singular que não é retratado na literatura, sendo necessário maior aprofundamento dessas diferentes manifestações no tegumento para evitar o diagnóstico médico errôneo e implementar o tratamento precocemente.

PALAVRAS-CHAVE: Hanseníase; Lesão linear; Dimorfa; Trajeto ulnar.

[1.] Universidade Federal do Pará, Núcleo de Medicina Tropical, Belém, PA. Brasil. ORCID: https://orcid.org/0000-0002-0611-5672. E-mail: nivinmsaid@gmail.com
[2.] Universidade Federal do Pará, Faculdade de Medicina, Belém, PA. Brasil. ORCID: https://orcid.org/0009-0002-7188-0984. E-mail: kenjimotizuk@gmail.com
[3.] Universidade Federal do Pará, Núcleo de Medicina Tropical, Belém, PA. Brasil. ORCID: https://orcid.org/0000-0002-5946-4876 . E-mail: Igorfroesm@gmail.com
[4.] Universidade Federal do Pará, Núcleo de Medicina Tropical, Belém, PA. Brasil. ORCID: https://orcid.org/0000-0003-2706-3880 . E-mail: mariliabxavier@gmail.com
Correspondence: Av. Generalíssimo Deodoro, 92 - Umarizal, Belém - PA, Brasil. CEP: 66055-240.

INTRODUCTION

Leprosy is a chronic neurodermatological infection caused by the etiologic agent *Mycobacterium leprae* (ML), an acid-fast bacillus that primarily affects peripheral nerves and the skin[1].

Brazil ranks second worldwide in absolute number of leprosy cases, behind only India[2,3]. Moreover, the disease is endemic in the country and constitutes a serious public-health problem due to its high potential to cause permanent physical disability and its persistently high endemic levels with varied distribution across different regions[2]. During the course of the disease and throughout treatment, leprosy reactions may occur; these are acute immunological episodes by which the host attempts to reduce the bacillary load, and they have an inflammatory character affecting both neural and dermatologic tissues[3,4]. Consequently, the clinical presentation of leprosy in the skin is variable and diffuse, since the disease expression depends on the immune response of each host to the bacillus.

CASE REPORT

A 17-year-old female patient from the interior of the state of Pará was referred to the tropical dermatology outpatient clinic. She reported a prior diagnosis of leprosy in her municipality and had received 8 doses of the multidrug therapy (MDT) regimen, but subsequently discontinued treatment on her own. Initially, she presented multiple hypopigmented, slightly erythematous, infiltrated plaques with poorly defined margins and irregular contours measuring approximately 20 cm in greatest diameter, as well as an ulnar claw deformity of the right upper limb (Figures 1 and 2). This ulnar claw represents a typical leprosy-related physical deformity and classifies the patient as having a grade 2 disability. After administration of the first dose of MDT the patient reported improvement of the skin lesions.

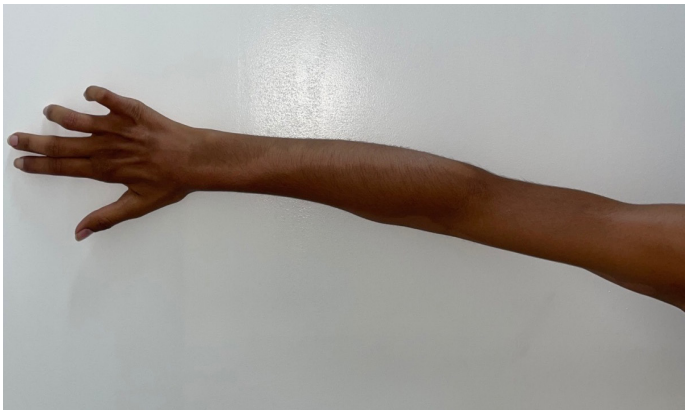

FIGURE 1 – Linear lesion on the posterior region of the right arm
Source: Authors

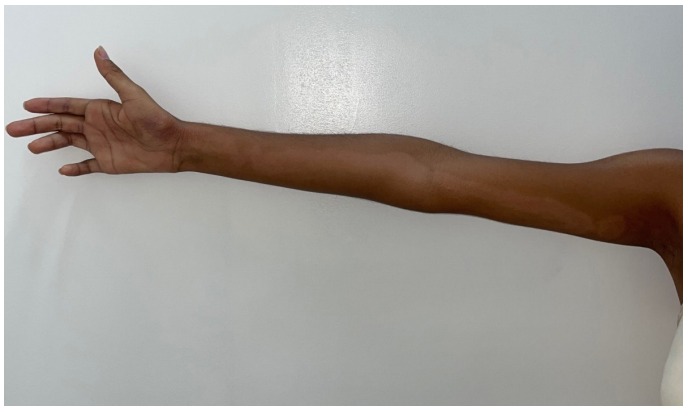

FIGURE 2 – Linear lesion on the anterior region of the right arm
Source: Authors

An incisional skin biopsy was performed for histopathological examination. Light microscopy revealed a lymphocytic inflammatory infiltrate, periadnexal fibrosis, and a perivascular and nerve-ending lymphohistiocytic infiltrate, indicating chronic dermatitis suggestive of leprosy. Correlating these histopathologic findings with the clinical data, the diagnosis was Bordeline–Bordeline leprosy (BB) (Figures 3 and 4).

During follow-up at the time of the second and third MDT doses, the patient reported a 30-day delay in menstruation and had a positive beta-hCG test. Cutaneous hypopigmented macules persisted on the arms, legs, and feet together with the ulnar claw of the right arm, which was associated with reduced strength of the fifth finger (ulnar nerve) and sensory loss in the ulnar distribution attributable to this deformity. She was advised to continue only MDT during the gestational period. The patient remains under infectious disease follow-up with ongoing guidance regarding management of physical disability, pregnancy in the context of leprosy, and scheduling of the next MDT dose.

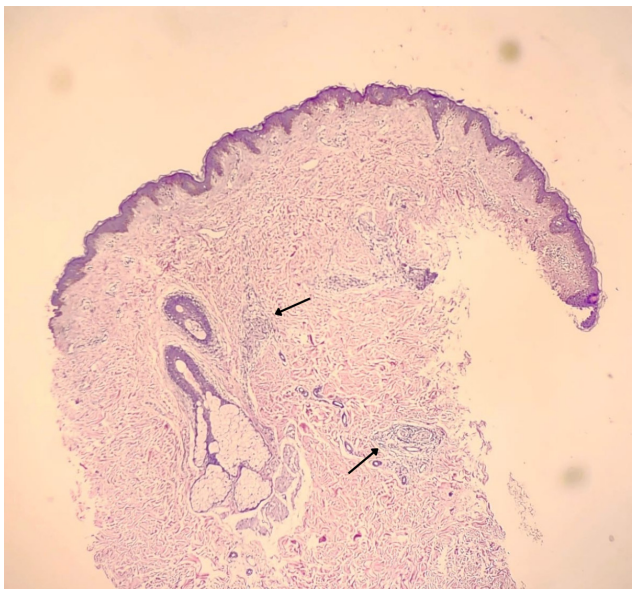

FIGURE 3 – Histopathological examination showing a lymphocytic inflammatory infiltrate, periadnexal fibrosis, and a perivascular and nerve-ending lymphohistiocytic infiltrate.
Black arrows indicate perineural granuloma
Source: Authors

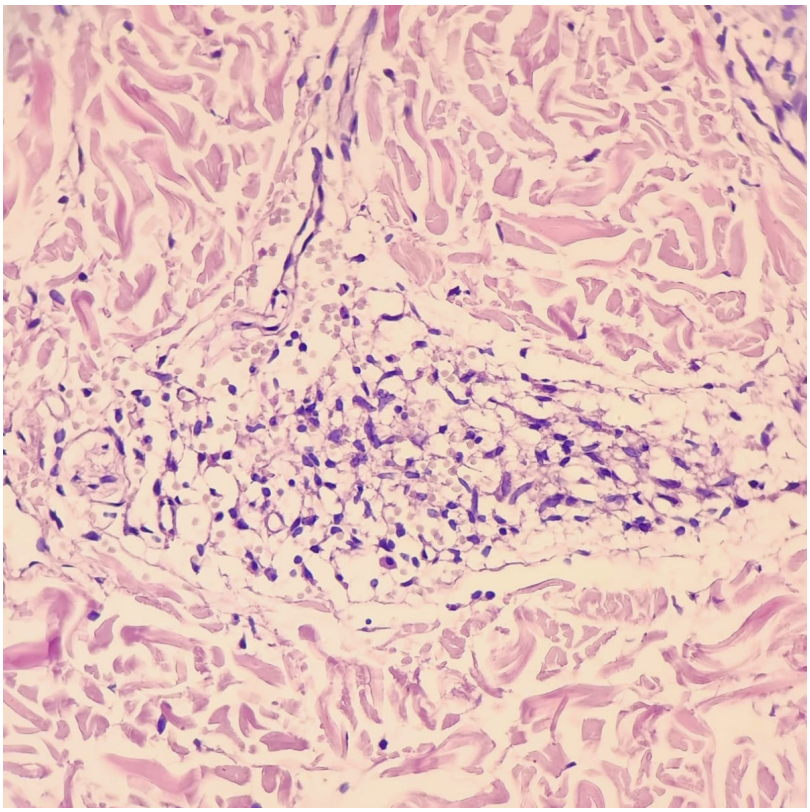

FIGURE 4 – Granulomatous infiltrate with histiocytes and plasma cells
Source: Authors

DISCUSSION

Leprosy belongs to the family Mycobacteriaceae and is historically documented as the first human disease to be attributed to a bacterial etiology. In addition, it is a disease capable of infecting a large number of individuals (high infectivity), although few develop clinical disease (low pathogenicity)[5]. Transmission occurs primarily from bacilliferous patients, the main source of infection, who expel significant numbers of bacilli via the respiratory tract; however, contagion requires direct and prolonged contact between healthy and infected individuals[6]. The bacilli multiply very slowly; the incubation period is typically two to five years and may reach up to 20 years, which complicates both epidemiological surveillance and early diagnosis[5,6].

Leprosy transmission is not restricted by social class or socioeconomic status, but its incidence is higher among the most vulnerable segments of society due to unfavorable economic conditions that may affect access to health services and housing, facilitating spread and endemicity[7]. In the present case the patient not only has unfavourable socioeconomic conditions but also resides in a locality with limited access to specialized leprosy care, requiring travel to the state capital to obtain multidisciplinary care, illustrating the ease of disease incidence among socially vulnerable populations. Given the coexistence of pregnancy and ulnar claw deformity in this patient, continuous monitoring is essential to ensure correct treatment follow-up.

Pregnancy has been associated with increased susceptibility to the onset or worsening of leprosy signs and symptoms, particularly between the last trimester and the first three months postpartum[8]. During gestation, women experience a relative suppression of physiological immunity— mainly cell-mediated immunity— predisposing to higher risk of infections, reactivations, or disease progression; therefore, frequent prenatal monitoring is required to assess both fetal growth and maternal health[9]. Studies by Ozturk and Tatliparmak (2017) and Helmer et al. (2004) report that MDT–PB can be associated with neonatal events, principally cutaneous manifestations such as clofazimine pigmentation of the skin,however there is insufficient scientific evidence to substantiate broad complications or long-term consequences in infants born to mothers treated with MDT[8,10]. The Brazilian Ministry of Health recommends treating pregnant women with the standard leprosy regimen (rifampicin, dapsone and clofazimine), advising caution particularly during the first trimester, since the benefits of treating maternal disease outweigh potential risks[8,11].

The clinical form of leprosy is determined by its spectral distribution associated with host immunological changes; the Ridley-Jopling classification is most commonly used clinically, relying on histopathological criteria and recognizing possible shifts along the spectrum toward the tuberculoid (T) or lepromatous (L) poles, with intermediate subtypes designated TT (tuberculoid), BT (borderline-tuberculoid), BB(borderline-borderline), BL (borderline-lepromatous) and LL (lepromatous)[12,13].

Numerous studies emphasize the variable cutaneous presentation of leprosy because M. leprae's cutaneous-neural manifestations depend on each host's immune response[13]. In

borderline patients and their subtypes, progressive reduction of immune response is accompanied by numerous skin and nerve lesions, increased bacillary load and higher antibody levels[13]. The DD form is considered immunologically dynamic, with frequent oscillation between the two polar forms[14].

Consequently, leprosy-related cutaneous manifestations vary in morphology, size and color according to each immune system's particularities, so that each lesion may appear uniquely on the integument, such as the linear-appearing lesion following the ulnar nerve trajectory seen in this patient. Nevertheless, leprosy retains characteristic dermatologic features detectable on exam, including erythematous-infiltrated plaques or hypopigmented patches[3,15]. Continued monitoring of uniquely appearing lesions is essential, since defining therapeutic measures to treat the extent or possible complications of such cutaneous manifestations during treatment is crucial to improve patients' quality of life and to highlight atypical integumentary presentations as possible differential diagnoses for leprosy.

Molho-Pessach and Schaffer (2011) emphasize that Blaschko's lines are products of cutaneous mosaicism—patterns of ectodermal clone migration during embryonic development—and may manifest as segmental or linear patterns of inflammatory and infectious diseases, including leprosy, suggesting that the patient described here might have epidermal cell groups genetically more susceptible to infection and thus more prone to lesions with this pattern[16,17]. Research by Chetan and Shailesh (2018) and by Liégeon and Reigneau (2015) supports the concept of locus minoris resistentiae, regions of lesser resistance to disease, which, together with Blaschko's lines, may identify areas of cutaneous mosaicism or nerve distribution that become points of reduced resistance to M. leprae and favor development of linear hypopigmented, erythematous or psoriasiform lesions[17,18].Therefore, genetic factors may also influence host immune responsiveness to the parasite.

CONCLUSION

The patient presented a linear cutaneous manifestation of leprosy on the right forearm, coinciding with ulnar nerve involvement manifested as an ulnar claw; the lesion followed the ulnar distribution. There is limited evidence in the literature, particularly in Brazilian publications, addressing the varied morphological or singular aspects of leprosy lesions beyond the Ridley-Jopling or Madrid classifications, which may lead to misdiagnosis or unnecessary testing and thus delay appropriate treatment. Continued and expanded studies that document the diverse clinical expressions of ML in humans are essential; a deeper understanding of the varied manifestations and associated features will facilitate earlier implementation of treatment and more accurate guidance concerning leprosy.

AUTHOR CONTRIBUTIONS: Nivin Said Mazen: case data collection, article selection, manuscript drafting, review, and formatting. Airton Kenji Motizuki: case data collection, article selection, manuscript drafting, review, and formatting. Nirlando Igor Fróes Miranda: case data collection, article selection, manuscript drafting, review, and formatting. Marília Brasil Xavier: supervision, guidance, review, and formatting.

REFERENCES

1. BRASIL. Ministério da Saúde. Secretaria de Vigilância em saúde. Boletim Epidemiológico: Hanseníase. https://www.gov.br/saude/pt-br/centrais-de-conteudo/publicacoes/boletins/epidemiologicos/especiais/2023/boletim_hanseniase-2023_internet_completo.pdf/view.
2. BRASIL. Guia de Vigilância em Saúde. Secretaria de Vigilância em Saúde. Brasília: Ministério da Saúde, 2014. p.294. https://bvsms.saude.gov.br/bvs/publicacoes/guia_vigilancia_saude_3ed.pdf.
3. Lastória JC, Abreu Mamm. Hanseníase:diagnóstico e tratamento. Diagn Tratamento. 2012;17(4):173-9. https://pesquisa.bvsalud.org/portal/resource/pt/lil-666961.
4. Pinheiro RO, Souza Salles J, Samo EM, Sampaio EP. Mycobacterium Leprae–Host-Cell Interactions and Genetic Determinants in Leprosy: An Overview. Future Microbiology. 2011;6(2):217-30. Doi: https://doi.org/10.2217/fmb.10.173.
5. Pires CAP, Chaves EC, Salmen CF, Balieiro ABR, Santos MBL, Araújo Filho GG, et al. Análise do Perfil Clínico- Epidemiológico da Hanseníase no Pará e Avaliação dos Indicadores de Saúde. Rev Eletr Acervo Saúde. 2019;27:e899. Doi: https://doi.org/10.25248/reas.e899.2019.
6. Azulay RD. Dermatologia. 8 ed. Rio de Janeiro: Guanabara Koogan; 2022.
7. Lopes VAS, Rangel EM. Hanseníase e Vulnerabilidade Social: uma Análise do Perfil Socioeconômico de Usuários em Tratamento Irregular. Saúde em Debate. 2014;38(103). Doi: https://doi.org/10.5935/0103-1104.20140074 .
8. Helmer KA, Fleischfresser I, Kucharski-Esmanhoto LD, Fillus Neto J, Santamaria JR. Fenômeno de Lúcio (Eritema Necrosante) na Gestação. An Bras Dermatol. 2004;79(2):205-10. Doi: https://doi.org/10.1590/s0365-05962004000200009 .
9. Amorim GM, Maestri T, Oppitz A, Leye M, Merini FL, Vignatti LJ. Hanseníase virchowiana e eritema nodoso hansênico em gestante de 34 semanas sem diagnóstico prévio. Rev Med, São Paulo, Brasil. 2022;101(1):e-170709. Doi: 10.11606/Issn1679-9836.V101i1e-170709.
10. Ozturk Z, Tatliparmak A. Leprosy treatment during pregnancyand breastfeeding: a case report and brief review of literature. Dermatol Ther. 2017;30:1-2. Doi: 10.1111/dth.12414.
11. Ministério da Saúde. Portaria n º 1073/GM de 26 de setembro de 2000. Publicada no D.O.U. - 100-E - página 18 - Seção 1 de 28 de setembro de 2000.
12. Ridley DS, Jopling WH. Classification of leprosy according to immunity: a five-group system. Int J Lepr Other Mycobact Dis. 1966;34:255-73. PMID: 5950347.
13. Mendonça VA, Costa RD, Melo G, Antunes CM, Teixaeira AL. Imunologia da Hanseníase. An Bras Dermatol. 2008;83(4): 343-50. Doi: https://doi.org/10.1590/S0365-05962008000400010.

14. Goulart IMB, Penna GO, Cunha G. Imunopatologia da hanseníase: a complexidade dos mecanismos da resposta imune do hospedeiro ao Mycobacterium leprae. Rev Soc Bras Med Trop [Internet]. 2002;35(4):363-75. Doi: https://doi.org/10.1590/S0037-86822002000400014.

15. Unterstell N, Machado PV, Obadia DL, Alves MFGS, Daxbacher E. Relato de Caso de Hanseníase Tuberculoide: Discussão Dos Achados Clínicos E Semióticos. Rev Hosp Univer Pedro Ernesto. 2011;10(1):24-9 . https://www.researchgate.net/publication/277712851_RELATO_DE_CASO_DE_HANSENIASE_TUBERCULOIDE_DISCUSSAO_DOS_ACHADOS_CLINICOS_E_SEMIOTICOS.

16. Molho-Pessach V, Schaffer JV. Blaschko lines and other patterns of cutaneous mosaicism. Clin Dermatol. 2011;29(2):205-25. Doi: 10.1016/j.clindermatol.2010.09.012

17. Chetan R, Shailesh M. Mid borderline leprosy in type Bα Blaschko linear pattern: a rare phenomenon. Int J Dermatol. 2019;58(6):729-32. Doi: 10.1111/ijd.14274.

18. Liegeon AL, Reigneau M, Rivail J, Bauvin O, Cuny JF, Schmutz JL. Lèpre tuberculoïde de disposition linéaire blaschkoïde: une présentation exceptionnelle [Linear tuberculoid leprosy along the lines of Blaschko: a very rare presentation]. Ann Dermatol Venereol. 2015;142(4):266-9. Doi: 10.1016/j.annder.2015.01.005.

Received: 2024, November 27
Accepted: 2025, August 14