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Case Report

A unique case of borderline lepromatous leprosy with psoriasis-like lesions all over the body and mycosis fungoides-like lesions on the face

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ABSTRACTS

Clinical manifestations of leprosy are various and may resemble other skin diseases. Skin lesions of leprosy mimicking psoriasis and mycosis fungoides (MF) that simultaneously occurs in one patient are rare. We reported a unique case of borderline lepromatous (BL) leprosy with severe reversal reaction manifested as psoriasis-like lesions and MF-like lesions in a 43-year-old-man. Psoriasis-like lesions all over the body accompanied by plaques and tumor-like lesions mimicking MF on the face could be found in this patient. Histopathological examination on an MF-like lesion from the face and psoriasis-like lesions from the posterior trunk and lower extremities revealed granulomatous reaction with epithelioid cells, Langhans giant cells, and foam cells which supported the diagnosis of BL leprosy. The patient was treated with multidrug therapy multibacillary (MDT-MB) regimen and 40 mg prednisone daily which was tapered off. Clinical improvement was observed on the 32nd day of observation as psoriasis-like and MF-like lesions became hyperpigmented macules and plaques, respectively. Due to the rarity of the multitype skin lesions of leprosy in one patient, a diagnosis of leprosy should be suspected by the clinicians in any patients with previously described skin disorders, especially in an endemic area.

1. Introduction

Leprosy is a chronic granulomatous infection caused by *Mycobacterium leprae (M. leprae.)* [1] In some cases, it can be caused by *Mycobacterium lepromatosis* [2,3]. Which mainly affects peripheral nerve tissue, skin [4], and can cause disabilities [1]. The clinical manifestation of leprosy might vary, ranging from macules (flat), infiltrates (elevated), or nodules [5]. The elevated skin lesions on leprosy might resemble other dermatoses, such as urticaria, lupus vulgaris, annular syphilis, sarcoidosis [5,6], or psoriasis vulgaris [5,7]. The nodular skin lesions can resemble cutaneous leishmaniasis, syphilis, cutaneous leukemia, or mycosis fungoides (MF) [5]. Several authors reported psoriasiform lesions on leprosy patients [7,8] and the others reported a leprosy case with clinical features resembling MF. However, histopathology of the skin lesions along with Fite's acid-fast staining revealed a large number of acid-fast bacilli (AFB) confirming the diagnosis of borderline lepromatous leprosy [9].

Reversal reactions might occur anytime on 30% of borderline leprosy (BL) patients with involvement of the skin, nerve tissue, or both [4]. In severe leprosy reactions, anti-reaction drugs should be administered immediately to prevent permanent disorder and disabilities [10]. The aim of this case report was to show leprosy as a "great imitator" disease, considering this disease might resemble psoriasis and MF in one patient.

2. Case reports

A 43-year-old male from a leprosy endemic area in West Java, Indonesia, presented with erythematous macules, plaques, and tumors all over the body and sometimes with itchy sensation since one month before admission. Approximately one year before admission, there was a non-pruritic and non-tender hypopigmented nummular macule on the left upper back of the body. Three months before admission, the patient noticed numbness with marble-sized tumors on the face (Fig. 1A and B) and erythematous plaques all over the body (Fig. 1C). The patient went to primary health care, then was diagnosed with leprosy, and received multidrug therapy-multibacillary (MDT-MB) regimen. One and half months later, the patient complained of cough and dyspnea, then decided to discontinue leprosy medication. Afterward, the patient was admitted to the district hospital with a diagnosis of pulmonary

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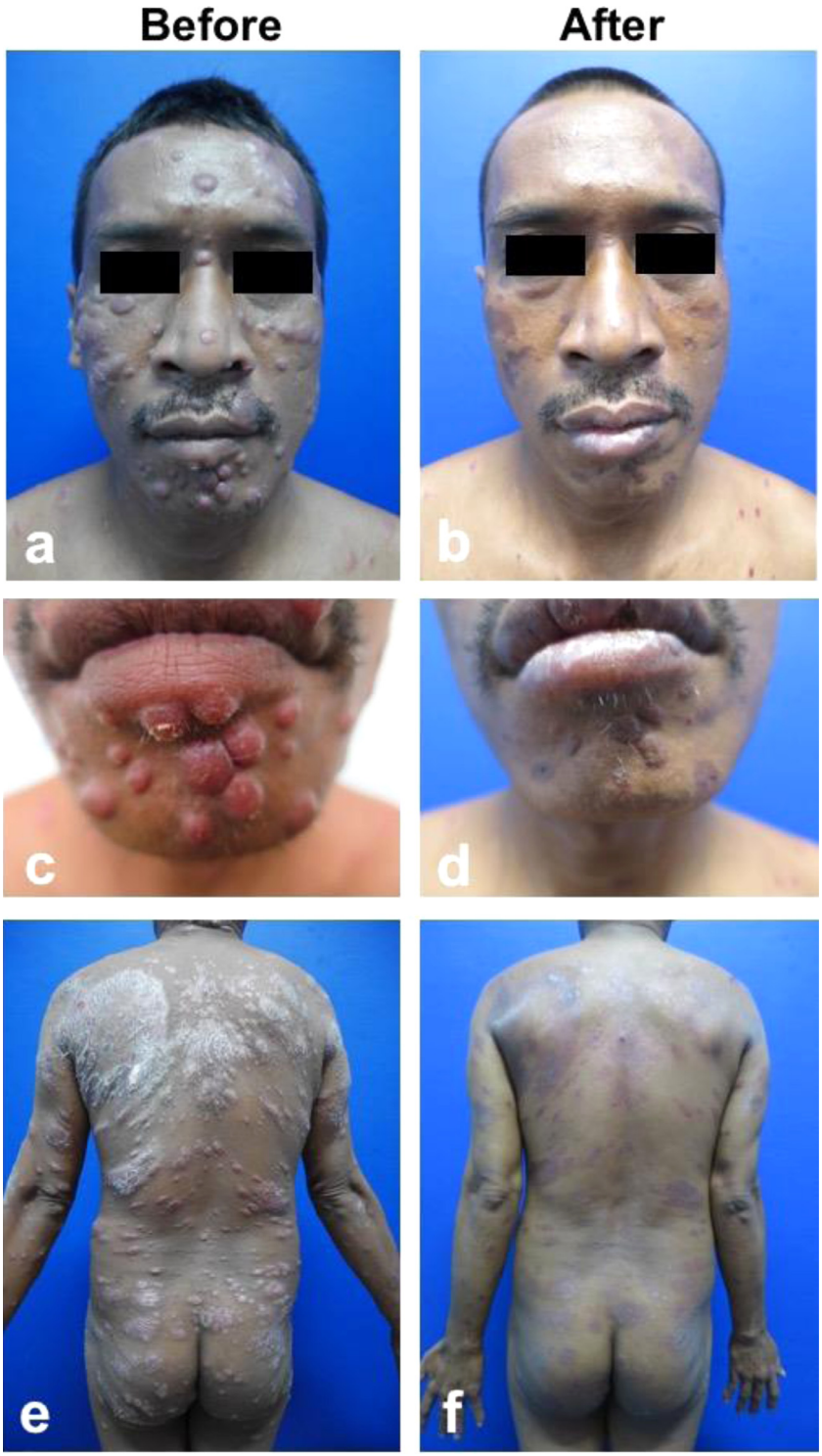

Fig. 1. Clinical manifestation of borderline leprosy (BL) leprosy. Before treatment: (a and c) Mycosis fungoides-like lesions on the face and (b) psoriasis-like lesions all over the body. After treatment: (b, d, and f) Significant improvement of the lesions after anti-tuberculosis treatment (ATT) and multidrug therapy- multibacillary (MDT-MB) treatment.

tuberculosis (TB), discharged with improvement, and was referred to our hospital to receive leprosy treatment again.

The physical examination showed edema on the upper and lower extremities. Hypesthetic erythematous papules and plaques covered with psoriasiform scales and crusts were found almost on the entire body, accompanied by plaques and tumors on the face. Meanwhile, psoriasiform scales with hypesthetic punched-out erythematous plaques were found on the left upper arm, abdomen, and left upper back. Non-tender enlargement of the right ulnar nerve and both common peroneal nerves were found with rubbery consistency and the left side of lower extremities stocking hypoesthesia. Thus, slit skin smear (SSS) examination revealed the presence of AFB with an average bacterial index (BI) of 0.75+ and morphological index (MI) 0% (Fig. 2A). Respiratory symptoms also occurred in this patient, then he referred to the Department of Internal Medicine and diagnosed with pulmonary TB and chronic obstructive pulmonary disease (COPD). Histopathological

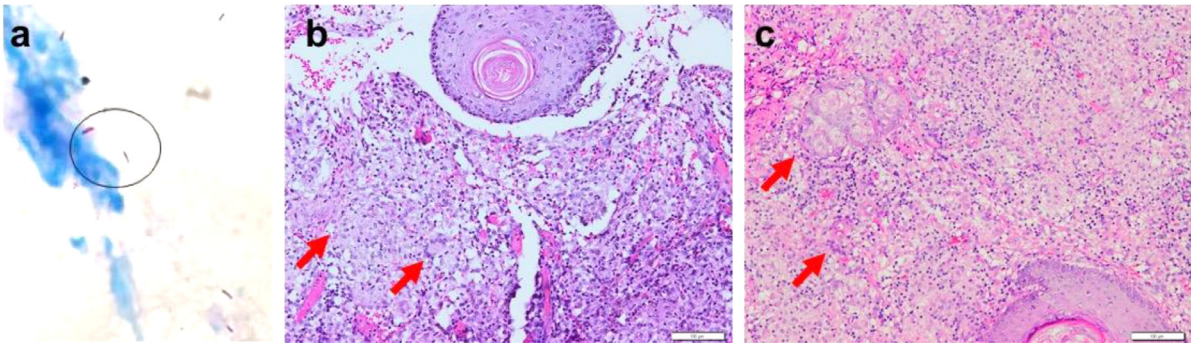

Fig. 2. (a) Acid fast bacilli (AFB) was found (in circle) from bacterial examination from left eyelid (Ziehl-Neelsen stain, ×1000 magnification). (b) Histopathological results of the psoriasis-like lesions on the left lower extremity and back showed Langhans giant cells (red asterisk) and foam cells (red arrow) (hematoxylin and eosin, ×100 magnification). (c) Histopathological results of the mycosis fungoides-like lesions on the face showed Langhans giant cells (red asterisk) and foam cells (red arrow) (hematoxylin and eosin, ×100 magnification). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

examination on an MF-like lesion from the face and psoriasis-like lesions from the left lower extremity and back showed features of granulomatous reaction with epithelioid cells, Langhans giant cells, and foam cells that supported the diagnosis of BL type of leprosy (Fig. 2B and C). Anti-phenolic glycolipid 1 (anti-PGL-1) immunoglobulin (Ig) M antibody level was 1270 μ/mL and IgG 422 μ/mL. The patient was treated with MDT-MB regimen for 12 months, 40 mg prednisone daily for the treatment of leprosy reaction, salbutamol, category 1 anti-tuberculosis treatment (ATT) that consists of isoniazid, rifampicin, pyrazinamide, and ethambutol daily for 2 months and followed by isoniazid and rifampicin. The improvement of skin lesions was seen on the 32nd day of observation (Fig. 1B, D, and F). For further evaluation, we could not confirm the effects after ATT and MDT-MB treatment, due to the patient moved to the other province and lost contact with the patient.

3. Discussion

Borderline leprosy is characterized by annular [5], “punched out”, or “swiss cheese” lesion with a clear inner border and unclear outer edge [4]. BL leprosy has many clinical presentations. It manifests in the skin as erythematous macules, papules, plaques, or nodules with the characteristic that lead to clinical diagnosis of BL [4,5]. One of leprosy clinical presentations could be erythematous plaques with clearly defined borders, accompanied with thick silvery scales, similar to the psoriasiform lesions in psoriasis vulgaris [1,7]. Psoriatic lesions in leprosy may manifest as erythematous plaques, which vary in size and shape. It primarily appears on the extensor area and also on the area which is prone to trauma, such as the knee, elbow, and buttocks. To distinguish psoriasiform lesions in leprosy from psoriasis vulgaris, there should be no loss of sensation, peripheral nerve thickening, AFB from SSS, and skin histopathological changes [11].

The clinical manifestations of MF are classified based on the clinical stages, ranging from the patch and the plaque stages (as the early stage) until the tumor stage (as the later stage). In the plaque stage, the skin lesions manifest as scaly plaques in red-purple color and some may appear to be indurated. The disease may remain in this stage or resolve spontaneously, leaving hyperpigmented lesions on the skin or progress to the severe form, the tumor stage. The cutaneous lesions of the tumor stage consist of papules and nodules in purplish red. The predilection sites of the lesion including the face, axilla, inguinal, infra-mammary fold, and forearm [9,12].

In this case, the patient showed hypesthetic erythematous papules and plaques covered with psoriasiform scales and crusts all over the body, with plaques and tumors on the face. Meanwhile, hypesthetic punched-out erythematous plaques with psoriasiform scales were found on the left upper arm, abdomen, and left upper back. There were three peripheral nerves thickening and AFB was found in the SSS examination. Based on these findings, the patient was diagnosed as leprosy.

Diagnosis of leprosy might be established in the presence of one or more of the cardinal signs, such as the appearance of hypopigmented or erythematous macules with sensory loss, peripheral nerve thickening with nerve function impairment, and presence of AFB on SSS [13]. Histopathology examination should be regarded as the gold standard in the diagnosis of a skin disease. It offers the higher sensitivity and specificity of the diagnosis and classification of leprosy, especially in patients with unclear clinical manifestation [11]. The histopathological examination result of this patient showed granulomatous reaction with epithelioid cells, Langhans giant cells, and foam cells that leads to the diagnosis of BL leprosy.

Phenolic glycolipid-1 is a specific surface antigen on the cell wall of *M. leprae*. These antigens can be detected through examination of urine, serum, nasal discharge, and skin biopsy specimen. This examination can help to determine the diagnosis and evaluation therapeutic response [14]. In this case, the result of anti-PGL-1 IgM was 1270 μ/ml (cut-off value: 605 μ/ml) and IgG was 422 μ/ml (cut-off value: 630 μ/ml), which confirmed the diagnosis of leprosy in this patient.

Leprosy reaction is the development of certain manifestation and acute inflammation episodes during chronic leprosy disease [13]. Based on clinical manifestation and histopathological findings, leprosy reactions are divided into type 1 and type 2 leprosy reaction [13,14]. Immunological response of this type of reaction may cause acute inflammation that affects the skin, peripheral nerve, or both. The predisposing factors of this reaction are chemotherapy, pregnancy, infection, physical, and emotional stress. The skin lesions become more erythematous and edematous, with peripheral nerve tenderness and impairment or loss of nerve function. Other physical findings are edema on certain areas of the body, such as the face, hands, feet, and also general state disturbances, such as fever and joint pain [13]. The patient on this case, there was edema on the upper and lower extremities that lead to the suspicion of reaction type 1 or reversal reaction.

The aim of the treatment of leprosy is to treat the acute inflammation sign and symptoms, reduce the pain, and prevent the upcoming nerve impairment. During the reaction state, MDT is necessary [6]. In mild reversal reaction, nonsteroidal anti-inflammatory (NSAID) drugs should be prescribed. The severe form of reversal reaction can be managed with systemic corticosteroid [15]. Systemic corticosteroid is the most common drug used for the treatment of leprosy reaction, due to its anti-inflammatory and immunosuppressive effect [4]. In general, the reversal reaction shows a good response to systemic corticosteroid therapy [15]. Based on World Health Organization (WHO) guidelines, the corticosteroid dose for early treatment is starting at 40 mg/day (prednisolone 40–60 mg/day), then gradually tapered off by 5–10 mg every 2 weeks, with a standard a 12-weeks course of treatment [16]. In

this patient, skin improvement occurred in 32nd day of observation, with a resolution of skin lesions. Leprosy reaction may also cause organ damage, such as kidney injury that may lead to death. Complications arising from nerve impairment are the most common cause of disability and deformity in leprosy patients [11].

4. Conclusion

This case report highlights a rare occurrence of multitype lesions of leprosy in one patient. The lesions appeared in psoriasis-like manifestation all over the body and MF-like lesions on the face. The histopathological examination can help to differentiate leprosy skin lesions from other skin diseases. Leprosy is “the great imitator”, therefore clinicians should raise suspicion of leprosy in any patients with skin disorder especially in an endemic area.

Consent and ethical clearance

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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CRediT authorship contribution statement

Hendra Gunawan: Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing. **Fitri Utami:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing. **Pati Aji Achdiat:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing. **Erda Avriyanti:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing. **Reti Hindritiani:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing. **Oki Suwarsa:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review & editing.

Declaration of competing interests

All authors of this research have no conflict of interest.

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References

[1] Agrawal US, Saxena A, Meena RS, Saini M. Hannsens's disease mimicking palmoplantar keratoderma. Int J of Leprosy 2002;70(3):210–2.
[2] Han XY, Seo YH, Sizer KC, Schoberle T, et al. A new mycobacterium species causing diffuse lepromatous leprosy. Am J Clin Pathol 2008;130:856–64.
[3] Han XY, Sizer KC, Vellarde-Fliex JS, Frias-Castro LO, Vargas-Ocampo F. The leprosy agents Mycobacterium lepromatosis and Mycobacterium leprae. Int J Dermatol 2012;51(8):952–9.
[4] Lee DJ, Rea TH, Leprosy MRL. Fitzpatrick’s dermatology in general medicine. In: Gogldsmith LA, Katz SI, editors. 8th edNew York: McGraw Hill; 2012. p. pg.2253–62.
[5] Yawalkar SJ. Leprosy for medical practioner and paramedical worker. 8th ed Switzserland: Novartis foundation; 2009. p. 51–9.
[6] Walker SL, Lockwood DNJ. The clinical and immunological features of leprosy. Br Med Bull 2006:77–8.
[7] Vora RV, Pilan AP, Jivan N, Kota RK. Leprosy mimicking psoriasis. J Clin Diagn Res 2015;9(9):1–2.
[8] Malin AS, Waters FR, Shehade SA, Roberta MM. Leprosy in reaction: a medical emergency. BMJ 1991;302:1324–6.
[9] Wang N, Lu X, Zang F. Follicutropic mycosis fungoides mimicking leprosy: a case report and review. Lepr Rev 2016;87:431–5.
[10] Kamath S, Vaccaro SA, Rea TH, Ochoa MT. Recognizing and managing the immunologic reactions in leprosy. J Am Acad Dermatol 2014;71(4):795–803.
[11] Sharma A, Sharma RK, Goswami KC, Bardwaj S. Clinicohistopathological correlation in leprosy. JK Sci 2008;10:120–3.
[12] Yamashita T, Marques EA, Abbade LPF, Marques SA. Mycosis fungoides and Sézary syndrome: clinical, histopathological and immunohistochemical review and update. An Bras Dermatol 2012;87(6):817–30.
[13] Bath RM, Prakash C. Leprosy: an overview of pathophysiology. Interdiscip Perspect Infect Dis 2012;2012:1–6.
[14] Lastoria JC, Abreu MAMMD. Leprosy: a review of laboratory and therapeutic aspects part 2. An Bras Dermatol 2014;89(3):389–403.
[15] Prasetya HY, Rahmah SN, Muchtar SV. Management of leprosy reaction. Ind J Dermatol Venerol 2012;4(5):24–34.
[16] World Health Organization. WHO model prescribing information: drugs used in leprosy. Geneva: World Health Organization; 1998.