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Case Report

Lucio's Phenomenon in Lepromatous Leprosy Patient: A Rare Case Report

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Abstract

Background : Lucio's phenomenon is a rare leprosy reaction, characterized by severe necrotic cutaneous lesions that generally occur in patients with untreated or inadequately treated lepromatous leprosy (LL). The objectives of this study was to describe and comprehend the diagnosis and management of Lucio's phenomenon in leprosy patients

Case : Male, 34 years old with extensive wounds on both arms and legs. About 3 years earlier, the patient felt burning heat in both hands and feet followed by loss of eyebrows and eyelashes, and a change in the shape of the nose. The patient had not received previous therapy. Physical examination showed that the patient appeared anaemic, leonine facies, megalobuli, madarosis, saddle nose, thickening of the ulnar and posterior tibial nerves, deformities of the fingers and toes, and amputations of several fingers. The dermatological status showed multiple ulcers of irregular shape and varying sizes with pus and necrotic tissue. Reitz serum test revealed a bacterial index of +3 with a morphological index of 90%, and routine blood showed hypochromic microcytic anaemia and leucocytosis. The patient received corticosteroid therapy, anti-leprosy combination therapy, antibiotics, iron tablets and wound care.

Discussion : Lucio's phenomenon consists of skin lesions that range from painful red-black patches to flaky blisters and ulcerations. These lesions usually occur on the lower extremities, and may extend proximally and distally. The lesions improved after administration of corticosteroids and anti-leprosy. Delayed diagnosis leads to significant disability and community transmission of the disease. The later the diagnosis, the more serious the disease becomes and can lead to death due to sepsis. Early diagnosis and appropriate treatment are important.

Conclusion : Early detection is crucial in order to deliver therapy earlier and prevent disease worsening.

Keywords : Lucio's phenomenon, Leprosy, Leprosy Reaction

INTRODUCTION

Lucio's phenomenon is a rare leprosy reaction. This phenomenon was first described by Lucio and Alvarado in Mexico in 1852, so it was called the Lucio's phenomenon. This condition often occurs in untreated or inadequately treated lepromatous leprosy. It can be life-threatening in some severe conditions. Lucio's phenomenon is very common in America, particularly in Mexico, although some cases have also been reported in Europe and Asia. Precipitating factors include pregnancy, stress, or infection. Lucio's phenomenon is difficult to diagnose, especially in non-endemic areas, causing delays in identifying the disease and starting time for treatment. Lucio's phenomenon is a type III hypersensitivity reaction associated with the deposition of immune complexes produced by antigen binding from the destruction of the bacilli with antibodies.[1–3]

The diagnosis is based on anamnesis, physical examination and Reitz serum and histopathology. Patient with Lucio's phenomenon usually presents with cutaneous ulceration in extremities. The symptom usually begins with redness that develop into ulcer and spread to the other parts of the body which not accompanied by fever and neuritis. It's also important to ask the patients if they had any contact with people who have leprosy or if they have ever been treated for leprosy.[1,3]

Lucio's phenomenon appears as dark red spots and becomes sores with necrotic tissue and associated deformities. These lesions will cause atrophic scars with a border of hyperpigmentation. Lesions are most often found on the hands, arms, legs and feet and rarely on the face and chest. One of supporting examinations for Lucio's phenomenon is skin slit smear examination. Examination of acid-fast bacilli (AFB) in the form of a skin slit smear stained with Ziehl-Nielsen staining taken from both ear lobes and skin lesion. Lucio's phenomenon showed a very positive bacterial and morphological index.[1,3]

In addition to skin slit smear examination, histopathology examination can also be performed. Histopathological finding shows colonization of endothelial cells by acid fast bacilli; endothelial proliferation of medium vessels of the mid-dermis with venous congestion; and neutrophil infiltration, ischemic epidermal necrosis, and necrotizing vasculitis of small vessels of the superficial dermis.[1,3,4]

The management of Lucio's phenomenon uses anti-leprosy regimen for multibacillary infection and systemic steroids. According to World Health Organisation (WHO) guidelines, multidrug therapy (MDT) for multibacillary leprosy was initiated for 18 months. High doses of corticosteroids (starting at 1 mg/kgBB equivalent to prednisone) should also be started and slowly tapered down over a period of months. After two weeks of treatment, the ulcers usually start healing. Bacterial and morphological index from skin slit smear examination are also performed to evaluate the treatment results. Management should be given at an early stage to reduce the occurrence of physical disability to death.[1,3,4]

CASE

A man, 34 years old, with the chief complaint of extensive and difficult to heal wounds on both forearms and both lower legs for 6 months ago. It was started as red-black spots which increased in number and became a wide and deep ulcer like a crater, with irregular edges, and painful. The ulcer occurred on both lower legs, then spread to the forearms to the fingers on both hands and feet (Figure 1). There was no fever and joint pain.

About 4 years ago, the patient complained of burning sensation in both arms and legs. The patient's eyebrows and eyelashes are falling out. The patient had never undergone treatment before. History of the similar complaints in the family was denied.

Physical examination showed anaemic conjunctiva, madarosis, megalobuli, *facies leonine, saddle nose*, deformities of the fingers and toes, and the amputate of the 4th and 5th fingers of the right and left hands. Nerve examination showed thickening of the ulnar nerve, posterior tibial nerve, and ulnar nerve paralysis. Dermatological status revealed hyperpigmented macules on the face, trunk and back, multiple ulcers on both arms and both legs with irregular shapes and varying sizes accompanied by pus, crusts and necrotic tissues (Figure1).

Complete blood investigation showed hypochromic microcytic anaemia (Hb: 5.3 mg/dL) and leucocytosis (15.35 x 103/ µL). Microbiological examination with Reitz serum on the earlobes showed a bacterial index of +3 and a morphological index of 90%. Whereas Reitz serum on skin lesions showed a bacterial index of +1 and a morphological index of 90%.

Based on anamnesis and physical examination, a diagnosis of Lepromatous Leprosy with Lucio's phenomenon was made. The management of this patient were rifampicin tablet 600 mg/2 weeks, clarithromycin tablet 500 mg/24 hours, methylprednisolone tablet 16 mg/2 hours, vitamin B complex tablet/12 hours, vitamin B12 tablet/8 hours, folic acid tablet/8 hours, iron tablet/8 hours, and zinc tablet 20 mg/24 hours. The treatment was given for a month then the patient was asked to control. Patients also received topical therapy in the form of 0.9% NaCl compress every 12 hours for 10 minutes, before applying cream to ulcers, 2% fusidic acid cream topical/12 hours on ulcers, and 1% silver sulfadiazine cream topical/12 hours on ulcers. After 30 days post treatment, the ulcer started to heal leaving a central achromic scar, surrounded by hyperpigmented

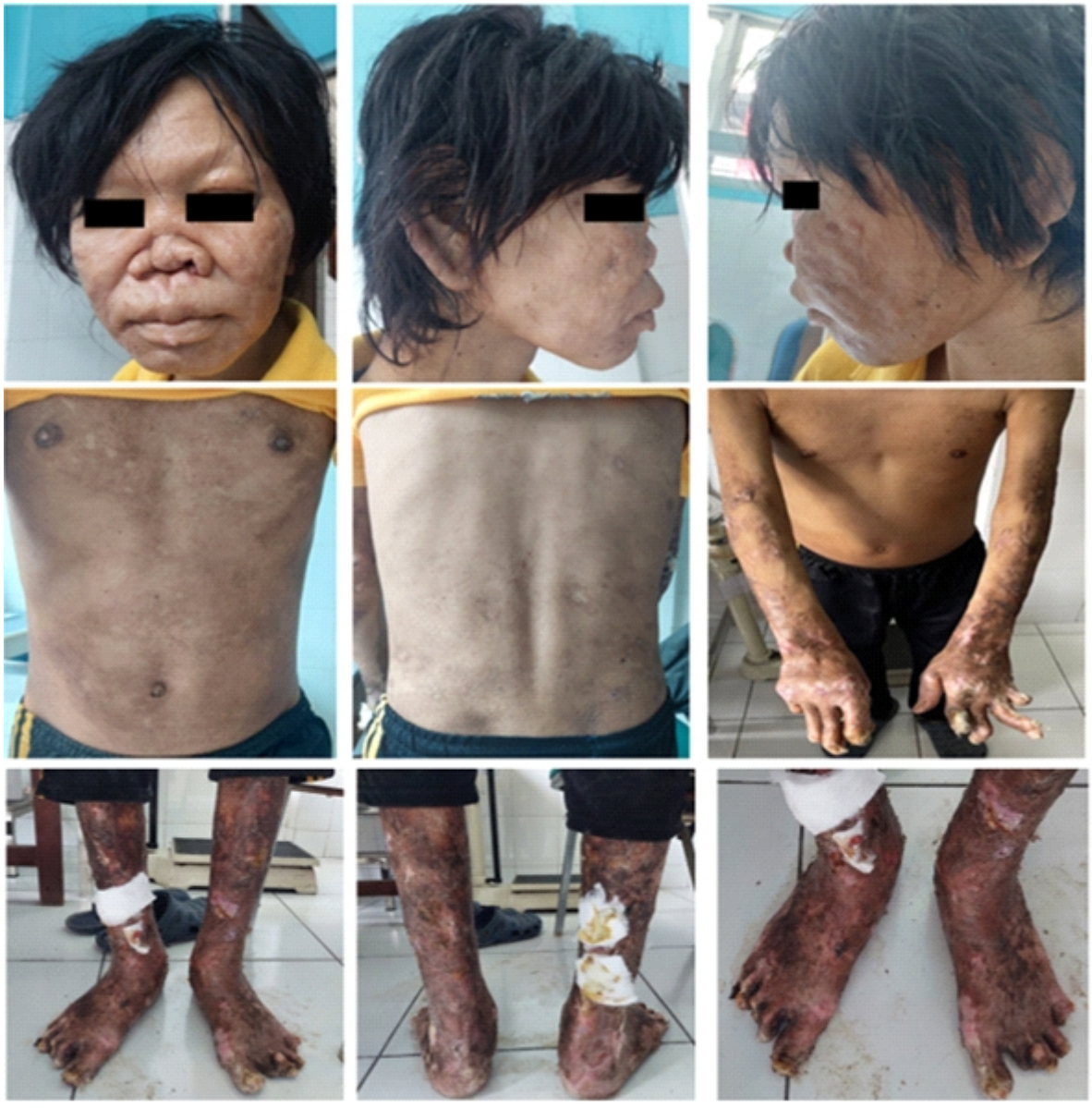

Figure 1. First day observation, before treatment

borders. Rifampicin and clarithromycin were continued for 18 months. After a month of administration, methylprednisolone was tapered off slowly according to the patient's clinical improvement.

DISCUSSION

The clinical manifestations of this patient match the diagnosis of Lepromatous Leprosy with Lucio's phenomenon. Lucio's phenomenon is a rare variant of the leprosy reaction, which is often found in the lepromatous version where it occurs due to unresponsiveness of the immune system causing unlimited proliferation of *Mycobacterium leprae*. As many as 0.7% of cases (16 cases) of a total of 843 cases of leprosy reactions in Indonesia. Lucio's phenomenon consists of skin lesions that range from painful red-black patches to flaky blisters and ulcerations.[5]

These lesions usually occur on the lower extremities, and may extend proximally and distally. The upper extremities, trunk and face may also be affected. Patients with Lucio's phenomenon usually do not have fever or neuritis.[1,6-9] Lucio's phenomenon usually occurs 1-3 years after disease onset and is common in patients with untreated or inadequately treated lepromatous leprosy and borderline type.[1,9-12]

The characteristics of lepromatous leprosy show the appearance of progressive skin thickening, thickened earlobes, rough facial lines and a concave shape. Facies leonine which may be accompanied by madarosis, iritis,

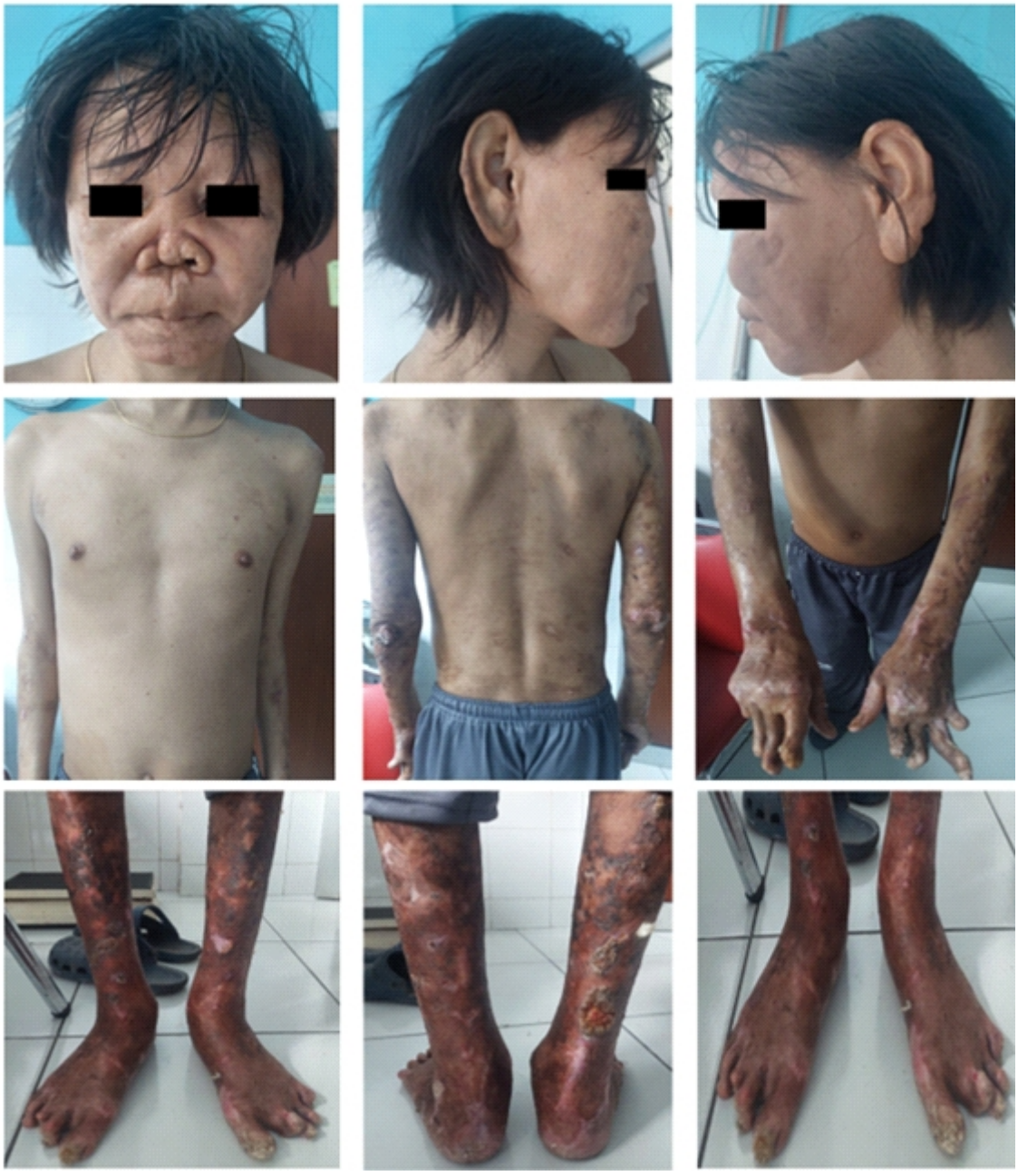

Figure 2. Observation day 30, after administration of anti-leprosy and corticosteroids

keratitis, and megalobuli. Furthermore, deformities can occur in the nose, called saddle nose. Peripheral nerve fibers experience thickening, hyaline degeneration, or fibrosis.[7,13]

Lucio's phenomenon begins as a bluish-red plaque, with a slight induration of the skin with an erythematous halo, usually on the extremities, but can also appear on other areas of the body. The lesions are ill-defined and painful with irregular or triangular shape. After a few days, the lesions will become purplish in the centre. Central haemorrhagic infarction may occur with or without blister formation. Subsequently, the lesion becomes a necrotic eschar, which detaches easily, and an ulcer with an irregular shape develops. The classic sign of Lucio's phenomenon is a jagged-edged necrotic ulcer with geometric shape.[7,9,10]

The results of supporting investigations in this patient are also consistent with the characteristics of Lucio's phenomenon in the literature. The Lucio's phenomenon characteristically shows very positive bacteriological and morphological index. The most common abnormalities found in laboratory tests are

anaemia, hypocalcaemia, hypoalbuminemia, leucocytosis, neutrophilia, and increased erythrocyte sedimentation rate. The classic clinical features of Lucio's phenomenon in this patient support the diagnosis of Lucio's phenomenon.[1,3]

Histopathological examination was performed in patients with atypical clinical features. Histopathological features will show colonization of endothelial cells by acid-fast bacilli, ischemic epidermal necrosis, necrotizing vasculitis of small vessels in the superficial dermis, endothelial proliferation of medium-sized vessels in the middle dermis with passive venous congestion and neutrophil infiltration.[9]

The principle of management in cases of Lucio's phenomenon is by administering high doses of corticosteroids equivalent to prednisone 1 mg/kg/day and starting anti-leprosy therapy. Rifampicin should not be given as monotherapy because resistance may develop. Rifampicin 600 mg or 1200 mg once a month was well tolerated. One of the new leprosy drugs, clarithromycin, is known to be very effective against infection *M. leprosy* in humans and has several advantages, namely providing clinical improvement, high bactericidal activity, good tolerance, and rarely causes side effects. Previous research stated that the combination regimen of rifampicin 600 mg per month and clarithromycin 500 mg per day for 3 months resulted in a significant reduction of acid-fast bacilli, a significant increase in healing, and no significant drug side effects were found.[1,9]

High doses of corticosteroids are tapered every month seeing clinical improvement in patients. Supportive care, such as wound care, is important in the management of these patients, to prevent secondary infection to sepsis. Plasmapheresis therapy can be an option if there is no improvement in the reaction.[13–15]

Evaluation of these patients showed a significant improvement. After 30 days post treatment, the ulcer started to heal and leave a central achromic scar, surrounded by hyperpigmented macules. In accordance with the literature that the ulcer will heal within a period of 2-4 weeks and leave a superficial atrophic scar.[7,10]

CONCLUSION

Lucio's phenomenon is a rare leprosy reaction that usually occurs in untreated leprosy. Early detection is needed to prevent morbidity and concomitant complications. This case was established based on clinical manifestations and investigations that support the diagnosis of Lucio's phenomenon. Administration of high-dose corticosteroid therapy and anti-leprosy showed clinical improvement in the patient.

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