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Default of Lepramatous Leprosy Patient with Lucio Phenomenon

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Abstract

Lucio phenomenon is type II leprosy reaction, The occurrence of Lucio's phenomenon is often associated with either no treatment or insufficient treatment for leprosy. We reported a case report default of lepramatous leprosy patient with lucio phenomenon. Lucio Phenomenon is difficult to diagnose. Some cases of Lucio's phenomenon are misdiagnosed. Early diagnosis and appropriate treatment of this case can reduce morbidity, mortality and the risk of complications.

Keywords : *Lucio Phenomenon, leprosy, default, necrosis*

INTRODUCTION

Leprosy reactions represent immunological phenomena that can manifest before, during, or after the completion of multidrug treatment (MDT). These reactions can be categorized into two types: type I and type II reactions. Lucio's phenomenon is classified as a type II reaction, typically observed in individuals with diffuse non-nodular lepromatous leprosy

or polar lepramatous leprosy (LL). The occurrence of Lucio's phenomenon is often associated with either no treatment or insufficient treatment for leprosy.[1,2,3]

CASE REPORT

A 22-year-old male patient presented with extensive wounds accompanied by discoloration on his hands and feet for the last 40 days. Initially, both legs displayed swelling and redness, progressing to a bluish hue resembling bruising and eventually evolving into blackened areas with widespread blistering. The patient did not report a specific onset tied to a skin lump. Additional complaints encompass sores on the buttocks, intermittent fever, weight loss, inability to walk, and numbness in the extremities.

At the age of 8, the patient received leprosy treatment at the Community Health Centre (Puskesmas) but discontinued the medication after just three months, ceasing treatment on his own. Six years ago, he began experiencing tingling and numbness in the soles of his feet, coupled with eyebrow hair loss. Both the patient's father and uncle underwent leprosy treatment and successfully completed the regimen.

Upon physical examination, the patient was found to be underweight, with a BMI of 17.8 kg/m^2. Dermatological abnormalities were noted in the bilateral auricular region, presenting as erythematous, diffuse nummular spots with scales, yellowish crusts, and areas of necrotic tissue (figure 1e). Madarosis, evident as eyebrow hair loss, was observed in the facial region (figure 1f). In the bilateral lower extremity region, multiple ulcers of varying shapes were present, characterized by non-elevated edges, absence of excavation, erythematous subcutaneous bases, and the presence of pus (+) and slough (+). Granulation tissue (-) was absent, and the most ulcers were covered with necrotic tissue. No nodes were observed (figures 1a and 1b). The bilateral upper extremity region displayed multiple ulcers of nummular-plaque size, with non-elevated edges and erythematous bases. Some ulcers had yellowish crusts, while others appeared blackish with necrotic tissue. Additionally, multiple hypopopigmented macules were present, circumscribed, and discrete in nature (figure 1c). The gluteus region exhibited multiple decubitus ulcers with flat edges, oozing, erythematous bases, firm borders, and nummular size (figure 1d). During the Range of Motion (ROM) examination, limited movement was noted in both the bilateral genu and bilateral cubital joints. Subsequent examination of the great auricular nerve revealed that the facial nerve functioned within normal limits. However, an assessment of other peripheral nerves was not performed due to the severity of the injury.

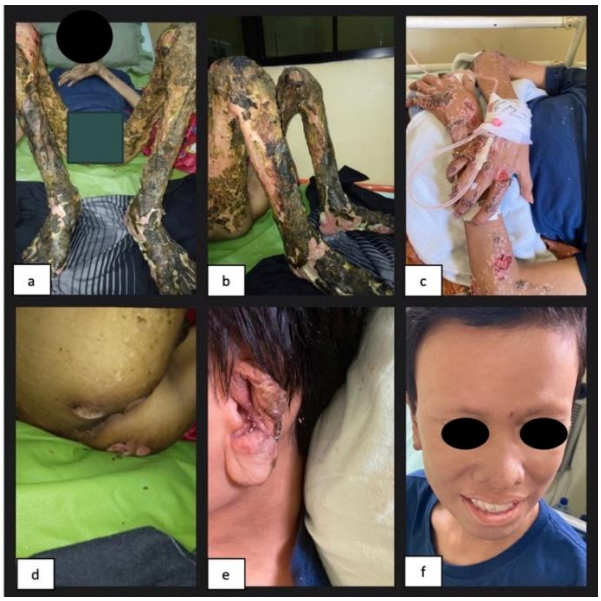

Figure 1 : (a) Inferior extremity region; **(b)** Inferior extremity region; **(c)** Superior extremity region; **(d)** Gluteus region; **(e)** Auricular region; **(f)** Facial region

The results of routine blood tests revealed a hemoglobin level of 6.6 g/dl, leukocyte count of 32.79 x 10^3/µL, erythrocyte count of 2.87 x 10^6/µL, hematocrit of 21,4%, and albumin level of 1.1 g/dl. Peripheral blood examination indicated hypochromic microcytic anemia.

A slit skin smear examination revealed the presence of acid-fast-bacilli (AFB) with a Bacterial index (IB) of 4.83 and an Infection Index (IM) of 1,33%. A complete stool examination showed the presence of spores (+) and hyphae (+). A histophatological examination with Hematoxyilin and Eosin (HE) staining on the patient's post-debridement skin tissue displayed tissue necrosis accompanied by groups of foamy histiocytes, distinctly demarcated from intact skin tissue. Vascular capillaries were obliterated with histiocyte infiltration dispersed among mononuclear inflammatory cells. A Ziehl-Neelsen (ZN) smear exhibited the presence of Virchow cells along with varied distributions of AFB, including globus, solid, and fragmented forms.

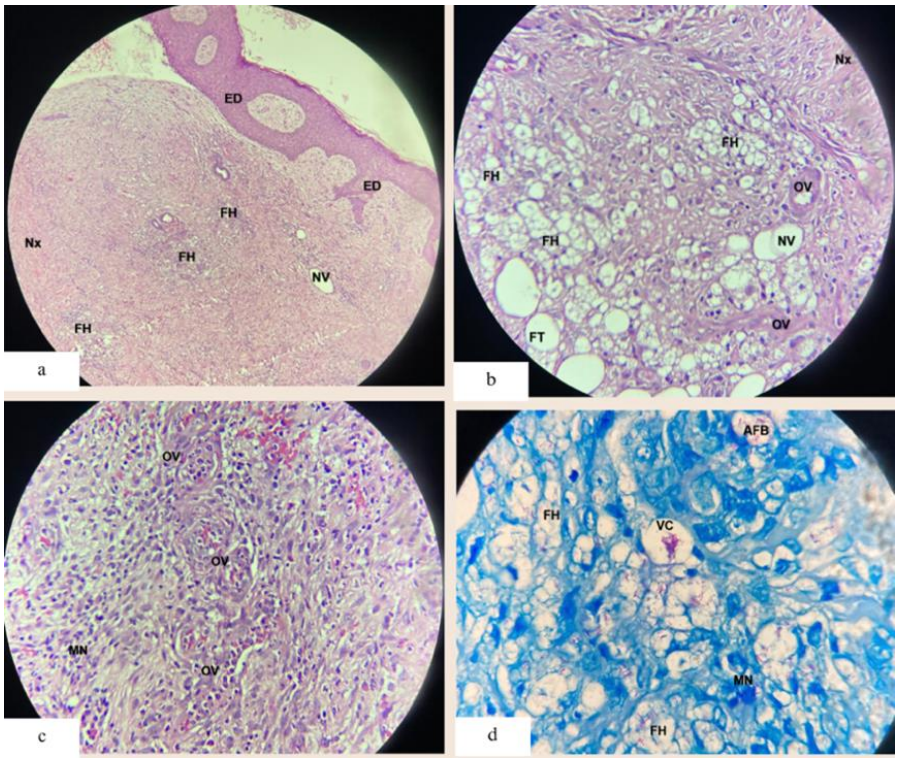

Figure 2 : (a) HE staining of histopathological biopsy of necrotomy tissue. Enlarged tissue (Nx) displaying groups of foamy histiocyte (FH) clearly demarcated by intact skin tissue (ED); **(b)** A 100x magnification displaying histiocyte infiltration in vascular vessel (NV) leading to vascular obliteration. Histiocyte infiltration extends into fatty tissue (FT); **(c)** A 400x magnification illustrarting the distribution of contaminated obliterated mononuclear (MN) inflammatory cells (OV); **(d)** A ZN staining depicting late phagocytosis of BTA (AFB) cells by histiocytes. Giant histiocytes are seen that form Virchow cells (VC).

The diagnosis of Lucio's Phenomenon was confirmed in a patient with Lepromatous leprosy, anemia, hypoalbuminemia, underweight, and intestinal mycosis. The patient received intravenous fluid therapy with Ringer's lactate at a rate of 20 drops per minute, methylprednisolone injection 2 x 31,25 mg, ceftriaxone injection 1 x 2 grams, metronidazole injection 500mg/ 8 hours. Additionally, the patient was administered rifampicin 1 x 600 mg, ofloxacin 1 x 400mg, minocycline 1 x 100mg, followed by Multidrug Therapy (MDT) MB. Other treatments included open compresses with NaCl 0,9%, mupirocin cream 2 x 1, prontosan, 1,000 cc packed red cell transfusion, albumin transfusion, albumin supplementation, fluconazole 1 x 150 mg, and a high-calorie protein diet. Extensive necrotomy was perfomed, and by the seventh day of treatment, the patient was deemed suitable for outpatient care. On July 18,2022, the patient was rehospitalized with debridement carried out on June 20,2022. On the sixth day of treatment, outpatient care was approved.

Figure 3 : (a) Dermatological status of the lower extremity region On July 7, 2022. Outpatient (D+1) after necrotomy; **(b)** Dermatological status of the lower extremity region on July 18, 2022; **(c)** Dermatological status of the lower extremity region on July 22, 2022 (D+2) after debridement. The ulcer base appeared cleaner.

DISCUSSION

Numerous studies conducted across various regions, including the United States, Peru, Hawaii, Brazil, India, Malaysia, and Indonesia, have linked Lucio's phenomenon to the *M.Lepramatous* species. [3,4] In the clinical context of this case, the initial presentation exhibited diffuse cutaneus infiltration without nodules, progressing to the development of reddish spots. If left untreated, these spots evolved into plaques and blisters, eventually transforming into extensive ulcers with necrotic tissue. The differential diagnosis for this case includes Erythema Nodusum Leprosum (ENL) reaction with vascular necrosis.

The table below delineates the distinctions between Lucio's phenomenon and ENL reaction with vascular necrosis. [5,6]

Lucio's phenomenon	**ENL reaction with vascular necrosis**
Occurs in leprosy with diffuse lesion without nodes/ papules/plaques, "*smooth rosy skin/ healthy appereance* " *****	Occurs in leprosy with plaque and node skin lesions
Occurs in patients who do not receive MDT treatment several years after disease onset, disappears with treatment, and sometimes changes shape into nodes *****	More often in the first month of MDT treatment
Reddish spot from, ulcers measuring 0,5-1 cm	Wide and deep necrotic ulcer lesions*
Hot sensation*	Ischemic pain
Usually afebrile*	Usually present with fever
Does not affect the nerves	Neuritis
No Constitutional symptoms or visceral damage	Artralgia, iridocyclitis, orchitis, lymphadenopathy,, nefritis, hepatitis
Tested positive using Medina	Tested negative using Medina
Histopathology : superficial leukocytoclastic vasculitis and necrosis, Macrophages containing BTA and proliferation of endhothelial cells in blood vessel walls*	Histopatology : superficial leukocytoclastic, deep leukocytoclastic, deep necrosis, erythema nodosum leprosum
Unresponsive to *thalidomide*	Responsive to *thalidomide*
Resolution in 15 days	Slower resolution*
Small scar tissue	Large scar tissue

*Clinical manefestations in the above case

The Pathophysiology of occlusive thrombi may result from immune reactions and the direct effects of Mycobacterium leprae within blood vessels.[7] The leading hypothesis suggests that bacterial liposaccharides stimulate macrophages to release TNF and interleukin (IL)-1. These products then act on endothelial cells, facilitating the production of prostaglandins, IL-6 , and coagulation factor III. [8]

The patient's histopathological examination with ZN smear revealed the presence of Virchow cells accompanied by the distribution of BTA. This aligns with the literature where Lucio Phenomenon demonstrated a significant number of BTA aggregates within the vascular endothelium. [9,10]

Diagnostic criteria for lucio's phenomenon encompass skin ulcers, vascular thrombosis, and Mycobacterium leprae invasion of blood vessel walls.[6] Establishing a diagnosis for Lucio's phenomenon is challenging, leading to some cases being misdiagnosed. [3] Early diagnosis and appropriate treatment can significantly reduce morbidity, mortality, and the risk of complications.

The patient, upon withdrawl from treatment, had only taken the MDT MB drug for 3 months and was declared in default due to a lack of family support and patient knowledge. Family support and supervision are essential for regular treatment, bit in this case, there was a

lack of it due to the patient's father's demise and the mother's frequent absence due to work commitments.

The patient, received rifampicin therapy 1 x 600 mg, ofloksasin 1 x 400 mg SD, minocycline 1 x 100 mg SD while awaiting MDT MB from the Puskesmas. There is currently no consensus on the treatment of Lucio's phenomenon, and existing are based on empirical studies and available case reports.[10] Numerous studies reporting of the effectiveness of the WHO MDT-MB regimen have shown its efficacy in treating Lucio's phenomenon.

Furthermore, the patient's condition included anemia, leukocytosis, hypoalbuminemia, being underweight, and severe mycosis, complicating the treatment process. Adressing the challenges requires a coordinated and collaborative approach from various medical fields, including surgery, internal medicine, medical rehabilitation, and nutrition.

The patient's treatment proceeded through outpatient care, focusing on education for rutine control, wound care, maintaining wound cleanliness, monitoring nutritional intake, and educating the patient on adhering to MDT MB medication to prevent repeated drug withdrawals.

CONCLUSION

We have reported a case of Lucio's phenomenon in patient with Lepromatous leprosy (LL) in a 22-year-old male. Clinical manifestations in patient with extensive wounds accompanied by discoloration on his hands and feet, on slit skin smear examination revealed the presence of acid-fast-bacilli (AFB) with IB of 4.83 and IM of 1,33%; the histopathological examinations found Virchow cells along with varied distributions of AFB. Management of the patient was given MDT MB, collaborative approach from various medical fields, including surgery, internal medicine, medical rehabilitation and nutrition. Necrotomy and debridement were carried out.

Lucio Phenomenon is difficult to diagnose. Some cases of Lucio's phenomenon are misdiagnosed. Early diagnosis and appropriate treatment of this case can reduce morbidity, mortality and the risk of complications.

REFERENCES

1. Vionni, Arifputra J, Arifputra Y. Reaksi lepra. CKD-242. 2016; 43(7): 501-4
2. Rocha RH, Emerich PS, Diniz LM, De Oliveira MBB, Cabral ANF, Do Amaral ACV. *Lucio's phenomenon: Exuberant case report and review of Brazilian cases*. An Bras Dermatol. 2016;91(5):60–3.

3. Widjaya SC, Mariam L. Diffuse Multibacillary leprosy with lucio's phenomenon in east Lombok, west Nusa Tenggara, Indonesia. International Journal of Dermatology, Venereology and Leprosy Sciences. 2021; 4(2): 25-8
4. Jurado F, Rodriguez O, Novales J, Navarrete G, Rodriguez M. Lucio's leprosy: a clinical and therapeutic challenge. Clin Dermatol. 2015;33(1):66e78
5. Fogagnolo L, de Souza EM, Cintra ML, and Ferreira Velho PE. Vasculonecrotic Reactions in Leprosy. Brazilian Journal of Infectious Disease. 2007;11(3):378-82.
6. Somani SP, Hemant HH, Milan M, Jhavar D, Astik BN, Lakhotia R. A vasculonecrotic type 2 Lepra Reaction Mimicking Lucio Phenomenin. JMID. 2019;9(3): 133-6
7. Pinheiro JV, Pontes MA, Neto JU, Goncalves HDS. Lucio Phenomenon: the importance of a primary dermatological care. Sociedade Brasileira de Dermatologia. 2022; 97(1): 54-7
8. Chauhan A, Palit A, Singal A, et al. IAL Textbook of Leprosy: Second Edition. 2017. p542–543.
9. Fung, W. K. Lepromatous Leprosy and Erythema Nodosum Leprosum. Hong Kong Dermatology & Venereology Bulletin. 2001. 10(1). Hal 28–30.
10. Frade MAC, Coltro PS, Filho FB, Horacio GS, Neto AA, Silva VZ, et al. Lucio's Phenomenon: A systematic literature review of definition, clinical features, histopathoenesis and management. Indian Journal of dermatology. 2022; 88: 464-77
11. Puspitasari, GD, Yuniaswan AP, Ekasari DP. A fatal case of lucio phenomenon in a patient with Lucio'Leprosy . JK-Risk. 2022; 1(3): 196-204