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Lucio's phenomenon

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Dear Editor,

A 32-year-old Indonesian female was a caregiver in Taiwan for 6 years. She had a 10-month history of intermittent pain in the bilateral elbows, hands, and knees. Based on a diagnosis of seronegative rheumatoid arthritis, she was treated with prednisolone (5 mg/day) and methotrexate (12.5 mg/week). One week before admission, she presented to a local hospital with diffuse purpuric macular lesions on her trunk, four limbs, and both ears. After four days of antibiotic treatment, the patient was referred to our hospital due to the progression of the disease. On admission, physical examination revealed normal body temperature, tachypnea (24/min), and sinus tachycardia (144 beats/min). Dermatological examination revealed retiform purpura with bullae and ulcerative lesions over the four limbs and trunk (Fig. 1A and 1B). A biopsy specimen of the left thigh skin was obtained on the second day of hospitalization. Histopathological analysis revealed thrombosis of small- and medium vessels, accompanied by infiltration of neutrophils, lymphocytes, and foamy macrophages distended with grayish-blue tinge globi in the dermis and fat lobules (Fig. 1C). Fite—Faraco staining revealed abundant acid-fast bacilli (Fig. 1D). A positive result of the *Mycobacterium leprae*-specific repetitive element (RLEP) real-time polymerase chain reaction for *M. leprae* led to the diagnosis of leprosy. The patient received multidrug therapy for leprosy (rifampicin 600 mg once monthly, dapsone 100 mg daily, and clofazimine 300 mg once monthly plus 100 mg daily), systemic glucocorticoids, and anticoagulation therapy. She also underwent surgical debridement and skin grafting for the wound. One month after the diagnosis, the patient died of massive duodenal bleeding complicated hypovolemic shock.

Leprosy is caused by *M. leprae*, or less commonly by *Mycobacterium lepromatosis*, which has an incubation period of approximately 2—20 years.[1,2] The disease primarily affects the peripheral cutaneous nerves, skin, and mucosa of the upper respiratory tract to the eyes, resulting in various clinical manifestations. Lucio's phenomenon is a rare manifestation in patients with lepromatous leprosy with rapid and threatening evolution. It is characterized by sudden erythematous-violaceous macular and slightly infiltrated plaques with cyanotic patterns on the four limbs but rarely on the trunk and face.[1] However, diagnosis is quite challenging, especially in non-endemic areas, leading to delayed disease recognition and appropriate treatment. Histopathologically, Lucio's phenomenon is characterized by a large number of acid-fast bacilli aggregates in the vascular endothelium and polymorphonuclear leukocyte inflammatory infiltrates in the deeper layers of the dermis and subcutis.[1] As the mechanism of Lucio's phenomenon is poorly understood, the most accepted hypothesis is uninhibited multiplications of bacilli and enhanced exposure of antigens to circulating antibodies, resulting in an immune complex occlusion in veins and subcutaneous tissue, progressing to a cutaneous hemorrhagic infarction.[1,3] The management of Lucio's phenomenon includes multidrug therapy, systemic steroids, immunosuppressants, and anticoagulants.[1,3,4] In these cases, exposure to immunosuppressive agents may result in high bacillary burdens. Despite the timely diagnosis and adequate treatment of Lucio's phenomenon in this patient, she died of other medical complications. Thorough history-taking to avoid misdiagnosis and incorrect treatment is the cornerstone of clinical practice.

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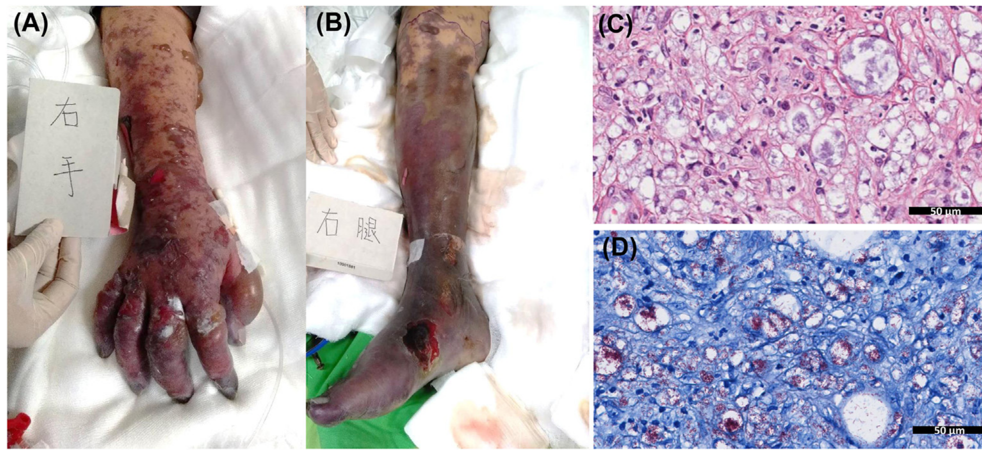

Fig. 1. (A) Multiple hemorrhagic bullae over right hand. (B) Diffuse, geographic, reddish to copper-colored purpura on the right leg. (C) Biopsy showing diffusely inflamed skin infiltrated with macrophages distended with large groups of bacilli. (D) A Fite-stain highlighting numerous acid-fast bacilli. The scale bar of each figure is shown in the lower right corner.

Ethics approval

The study has been approved by the Institutional Review Board of Tri-Service General Hospital (TSGHIRB No.:C202215010) and granted the waiver of informed consent.

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None.

Declaration of competing interest

There are no conflicts of interest.

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