

Case report

Lucio phenomenon – a rare life threatening state of leprosy

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Abstract

The Lucio phenomenon is a highly uncommon and severe form of reactional state associated with leprosy, particularly occurring in patients with untreated diffuse, non-nodular lepromatous leprosy. It manifests as a necrotising form of extensive cutaneous vasculitis, resulting in necrotic and haemorrhagic ulcers on the extremities. Despite its rarity and significant mortality rate, only a limited number of cases have been reported from Asian countries.

We present a case of a 56-year-old male diagnosed with lepromatous leprosy and receiving the WHO-recommended multidrug treatment regimen. The patient presented with extensive nodulo-ulcerative lesions and minimal systemic symptoms. Histopathological examination confirmed the diagnosis of Lucio phenomenon rather than ulcerated erythema nodosum leprosum, a condition that mimics Lucio phenomenon. Notably, the patient exhibited a high morphological index (MI) and bacillary index (BI) in slit skin smear, despite receiving the recommended duration of one year of multibacillary treatment.

Intravenous immunoglobulin (IVIG) was administered to the patient, resulting in complete re-epithelialization of the ulcers. This case underscores the importance of high clinical suspicion in diagnosing Lucio phenomenon and the timely use of IVIG to halt the progression of extensive vasculitis, thereby minimising complications.

Key words: Lucio phenomena, leprosy, necrosis, ulcerations, immunoglobulin

Introduction

Leprosy is a chronic infectious disease caused by *Mycobacterium leprae*, that may be complicated by acute immunological lepra reactions. These lepra reactions may occur before, during, or after the end of anti leprosy treatment. Lucio phenomenon is a very rare and severe form of reactional state of leprosy, peculiarly occurring in patients with untreated diffuse, non nodular form of lepromatous leprosy. It is commonly found in Mexico, but it occurs very rarely in the endemic areas of the rest of the world. It is a

necrotising form of extensive cutaneous vasculitis leading to necrotic and haemorrhagic ulcers on the extremities. Failure to recognise this Lucio phenomenon and delay in appropriate treatment may lead to significant morbidity and even mortality in these leprosy patients.

Case history

A 56 year old male presented to the dermatology unit of the Teaching Hospital, Batticaloa with extensive ulcerated lesions over both upper and lower limbs for one month duration. He was being treated with the standard multibacillary regimen, after being diagnosed as lepromatous leprosy with grade two disability in a base hospital one year ago. At the initial diagnosis, highest values of bacillary index (BI) and morphological index (MI) for *Lepra* bacilli were 4+ and 65% respectively in this patient. Four months after the initiation of treatment, he developed symptomatic anaemia with evidence of haemolysis, most likely induced by dapsone. Since then, the MDT-MB treatment regimen was continued without dapsone. After completion of 12 month MDT-MB treatment, he dramatically developed extensive necrotic ulcerations over both upper and lower limbs. The onset of ulcers was preceded by painful purpuric plaques and nodules which appeared in a more generalised distribution involving face, trunk and limbs. These lesions were ulcerated only over the limbs. These acute cutaneous eruptions were associated with minimal systemic symptoms. At time of admission, the patient was found to have characteristic leonine facies including infiltrated facial skin, madarosis, saddle nose deformity and nodular thickening of ear lobes (Figure 1). There were extensive punched out shallow ulcerations of varying sizes mainly confined to upper and lower limbs (Figure 3 & 4). He was also noted to have clawing in hands and feet. There were non ulcerated purpuric plaques over thighs, trunk and face (Figure 2).

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Figure 1. Characteristic leonine face in lepromatous leprosy.



Figure 2. Purpuric plaques.



Figure 3.



Figure 4.

Figure 3 and 4. Necrotic and haemorrhagic bizarre shaped punched out ulcers involving lower limbs and upper limbs.

He was extensively investigated for the condition. Repeated highest BI and MI values were 3+ and 15% respectively. He was noted to have anaemia of chronic disease and severe hypo-albuminaemia (serum albumin 9g/dl). Despite the above findings, his liver biochemistry was normal other than high Gamma-GT level which was explained by his chronic alcoholism. ESR and CRP were very high. Skin biopsy was performed and the histology revealed extensive areas of necrosis with focal vasculitis and the viable skin with granulomas, concluded as lepromatous

leprosy with necrotising vasculitis (Figure 5 & 6). Wade-Fite stain revealed lepra bacilli invading the vessel wall.

He was initially started on IV immunoglobulin 2g/kg body weight in divided days and clofazimine 100mg tds to control the severe lepra reaction. As his serum procalcitonin level was very high indicating underlying severe sepsis, systemic steroid was not started. His sepsis was managed with IV meropenem and teicoplanin.

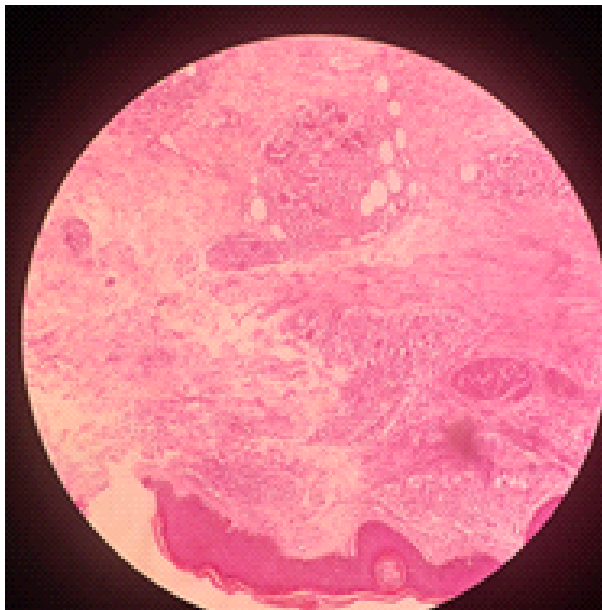


Figure 5. (Histology) viable skin with granuloma formation.

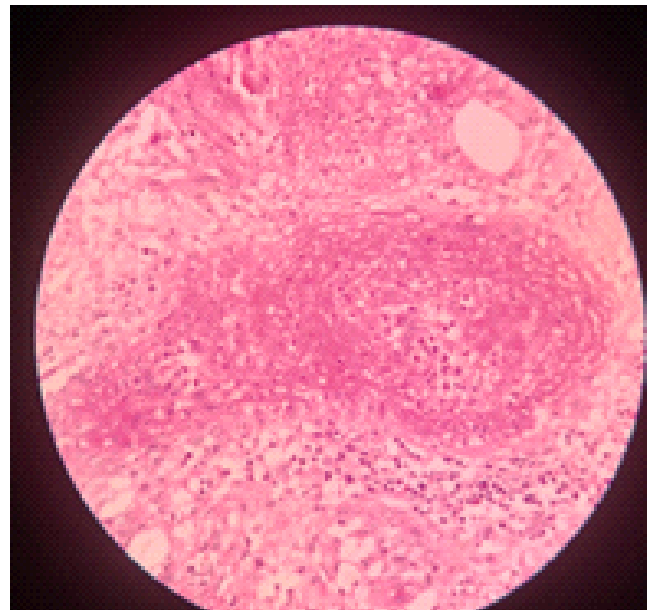


Figure 6. (Histology) Focal necrotising vasculitis.

By the 3rd week after IV immunoglobulin, patient was noted to have significant improvement in cutaneous ulcerations with granulation tissue formation and re-epithelialization (Figure 7 & 8).



Figure 7.



Figure 8.

Figure 7 and 8. Necrotic and haemorrhagic bizarre shaped punched out ulcers involving lower limbs and upper limbs have almost completely reepithelialized – 3 weeks after IV immunoglobulin.

Discussion

Lucio phenomenon is a rare, aggressive, ulcerative form of type 2 lepra reaction. It develops due to exacerbated proliferation of lepra bacilli invading the walls of blood vessels and endothelial cells leading to thrombosis within the vascular lumen resulting in ischaemia followed by necrosis of tissues. To diagnose as Lucio phenomenon three criteria are to be met: skin ulceration, vascular thrombosis and invasion of blood vessels by the leprosy bacilli. Clinically, it presents as multiple purpuric plaques and macules that evolve into massive bizarre shaped ulcerations on extremities, healing with atrophic scarring. Fever and constitutional symptoms are minimal. It is important to differentiate it from ulcerated form of erythema nodosum leprosum which usually presents with erythematous painful nodules followed by ulceration and marked constitutional symptoms. Starting IV immunoglobulin at the early stage will halt the immune reaction. It promotes reepithelialization of ulcers early. Thalidomide and clofazimine are less effective in Lucio phenomena as compared to erythema nodosum leprosum. We report this case as Lucio phenomenon in leprosy is extremely rare in our region and IV immunoglobulin proved to be the best treatment option in healing extensive cutaneous ulcers dramatically.

Conclusion

A high degree of clinical suspicion and characteristic histology is needed to diagnose Lucio phenomenon, a rare form of lepra reaction. Histopathological findings play a key role in differentiating it from ulcerated ENL. In a setting where prednisolone is contraindicated, early starting of IV immunoglobulin could yield a remarkable treatment outcome in the management of Lucio type of severe lepra reaction.

References

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