

Polymorphic light eruption-like lesions in association with cutaneous leishmaniasis – is it a unique leishmanid reaction in *Leishmania donovani* cutaneous leishmaniasis in Sri Lanka?

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Abstract

Cutaneous leishmaniasis (CL) in Sri Lanka is caused by *Leishmania donovani* species, which usually causes visceral leishmaniasis in neighboring countries. We report a case series of 54 patients with cutaneous leishmaniasis from Anuradhapura, Sri Lanka, with associated polymorphic light eruption (PMLE) like lesions. As these PMLE-like lesions show characteristic features of an id reaction, we propose that this could be a leishmanid reaction. To date, PMLE-like leishmanid reactions are not reported in association with other leishmania species. Therefore, we suspect that PMLE-like leishmanid reaction could be a unique association of *Leishmania donovani* CL that occur in Sri Lanka.

Case series

This case series was collected over a period of one year at the Teaching Hospital, Anuradhapura, Sri Lanka. This consist of 54 patients with cutaneous leishmaniasis with an associated skin eruption in photo-exposed areas.

The majority were males (87.5%) and had a single CL lesion (88.8%). All patients were systemically well with no hepatosplenomegaly. The clinical diagnosis of CL was confirmed either parasitologically with smears or histologically. The average duration of the CL before presenting to treatment was 6.6 months. The associated skin lesions on photo-exposed areas were clinically suggestive of PMLE. Different clinical morphologies of the PMLE-like eruption, such as hypopigmented macules, grouped flat-topped papules and scaly plaques were noted (Figure 1-3). Out of these, hypopigmented macules were the commonest (74%). The PMLE like eruption affected the upper limbs in the majority (92%). The average duration of PMLE-like lesions was 2.7 months at presentation. Fourteen patients had received topical steroids for the PMLE like lesions before coming to the dermatology clinic. Others had not received any medication including ayurvedic medications, earlier.

Skin smears and biopsies for histology were done from PMLE like eruptions in 30 consenting patients. The CL lesions were treated with weekly 1-3ml of intra lesional sodium stibogluconate depending on the size of the lesion. No additional treatment was offered for the PMLE like eruption.



Figure 1. Grouped hypopigmented papules.



Figure 2. Hypopigmented macules.

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Figure 3. Scaly plaques with both hypo and hyperpigmentation.

Twenty-eight biopsies showed evidence of PMLE such as spongiosis, mild basal cell vacuolation and acanthosis of the epidermis, and edema and perivascular lymphocytes infiltrate in the superficial dermis (Figure 4). Occasional dyskeratotic cells, lymphocytes exocytosis and periadenexal lymphocytes infiltrate were also seen. The other two showed non specific chronic inflammation. None showed features of leishmaniasis parasitologically or histologically, excluding a diagnosis of an atypical presentation of CL or post-Kalar azar dermal leishmaniasis.

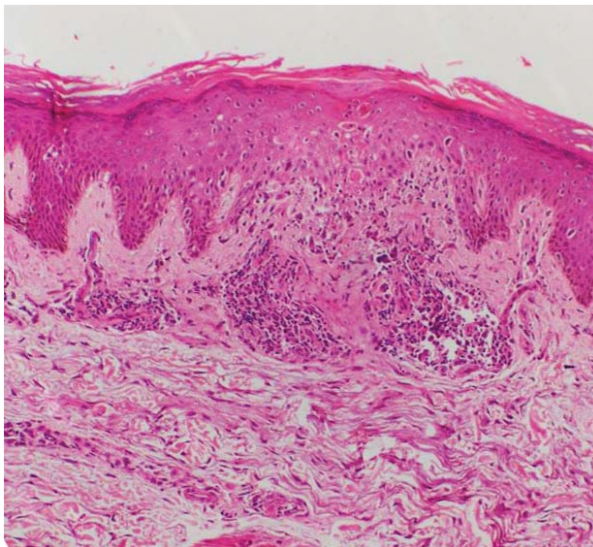


Figure 4. H&E, × 100. Skin biopsy showing epidermal acanthosis, spongiosis and scattered dyskeratotic cells. The papillary dermis shows oedema and lymphocytes infiltrate.

50 out of 54 (92.5%) showed complete remission of their PMLE-like lesions without any steroid treatment in an average of one month, with a range of 3 weeks to 2 months (Figure 5a and 5b). Four patients showed worsening of PMLE like eruption with the initiation

of SSG. However, the PMLE like eruption achieved complete clearance before the CL was fully cured in all patients. Furthermore, fourteen of this series had received topical steroids for PMLE-like lesions before the diagnosis of CL without much response. Even in this subset, it was observed that PMLE-like lesions disappeared following treatment of CL with intralesional SSG.



Figure 5a. PMLE like lesions in a 36 year old male with ulcerative CL on left forearm.



Figure 5b. Complete resolution of PMLE like lesions and CL after 4 weeks of intralesional SSG.

Discussion

Cutaneous leishmaniasis (CL) in Sri Lanka is caused by *Leishmania donovani* species¹. It is now an established endemic disease in the country.

Id reaction is a non-specific skin reaction which occurs as an immunologic reaction to an infective or inflammatory focus². This most commonly occurs with a superficial fungal infection (dermatophytid). The clinical features of id reactions are mostly papular vesicular lesions. However, nodular, eczematous, urticarial, scarlatiniform, and bullous lesions are reported².

Leishmanid reaction was first reported by Berlin *et al* in 1940³. Leishmanid reaction by itself is a rare occurrence and different morphological patterns are described^{3,4}.

Id reaction can occur with infective foci or with an inflammatory skin disease. Usually, this is a secondary immunological reaction of circulating antibodies or activated T lymphocytes directed against infective antigens. This usually occurs at the height of infection, shortly thereafter or immediately after starting treatment due to high antigen release.

Id reaction shares the following characteristics: (i) the host harbors a proven focus of infection elsewhere; (ii) a positive intradermal skin test (iii) infective forms are neither recoverable from the site of the cutaneous eruption nor microscopically visible in direct smears or histopathologic preparations; and (iv) the lesions spontaneously resolve after an acute course, provided that the primary infection is identified and eliminated². Four out of three criteria were fulfilled by all our patients indicating that the rash that occurred in photo-exposed areas was an id reaction. The leishmanin test was not done due to unavailability.

Morphology of id reaction can vary considerably. Different morphological types of leishmanid reactions are described⁴. To the best of our knowledge, a PMLE-like leishmanid reaction is not reported to date. Our patients' skin lesions associated with CL were consistent with PMLE clinically.

Histology of id reactions is non-specific and will be consistent with the clinical variety of the same². As such our patients' histology was inconsistent with PMLE with no indication of leishmaniasis.

By definition, id reactions resolve spontaneously when the primary infective foci are eliminated. Oral

steroids are offered as a symptomatic treatment for severe id reactions. However, id reactions can be quite unresponsive to steroids³. The PMLE like eruption in all patients' subsided when the CL lesion was treated with intralesional SSG, further pointing towards a leishmanid reaction.

One might argue that these lesions are true PMLE, which occurred concurrently with CL, as the majority of our patients are outdoor workers. PMLE usually occurs in females and in this case series 87.5% were males. PMLE typically describe acute skin lesions, which occurs within hours or a few days of sun exposure⁵. None of our patients could recall a certain day of intense sun exposure before the onset of the rash and the onset was more insidious. Additionally, a subset of the series showed unresponsiveness to topical steroids, unlike true PMLE. Considering these it is more likely that the PMLE-like lesions are a true association of CL, more specifically a leishmanid reaction rather than a mere coincidental finding.

This case series highlights a large collection of patients with CL with associated PMLE-like skin lesions which could be a leishmanid reaction. The causative species of CL is unique to Sri Lanka. The morphological variety of PMLE-like leishmanid reactions is not described in the literature. Therefore, there is a possibility that this is a unique association of *Leishmania donovani* CL. Our case series is a clinical observation and properly designed clinical studies will be needed to establish this disease entity.

References

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