

# Unusual clinical presentation of Cutaneous Leishmaniasis in a patient with corticosteroid induced immunosuppression

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## Abstract

Cutaneous Leishmaniasis (CL) is often a localized and self-limited disease, but its behavior changes in the state of immunosuppression. Here, we report a rare clinical presentation of disseminated CL after reactivation of leishmania infection in a 42 year old male with corticosteroid induced immune suppression.

## Keywords:

Cutaneous Leishmaniasis, Disseminated, Immunosuppression, Systemic Corticosteroids, Recurrence

## Key Clinical Message:

- Immunosuppressive treatments alter the clinical manifestations of Cutaneous Leishmaniasis
- Unconventional manifestations can cause a delay in diagnosis
- Diagnosis needs further evaluations and special consideration in immunosuppressed patients

## Introduction

Cutaneous Leishmaniasis (CL) is a parasitic infectious disease which is endemic in several countries around the world including Iran. This infection is caused by a group of protozoan parasites of the genus *Leishmania* and is transmitted by the bite of infected female phlebotomine sandflies. (1)

In Iran CL is mostly caused by *Leishmania.major* and *Leishmania.tropica* species. (2) The clinical features of CL depend on various factors including *Leishmania* species, environmental factors, genetic factors and immune response of the host. (2) Innate and adaptive immune responses play an important part in controlling leishmania infection and immunosuppression is a known risk factor for unusual clinical variants of CL. (3, 4)

This case report highlights the unconventional clinical manifestation and reactivation of CL in a patient undergoing systemic steroid therapy and also demonstrates that the diagnosis could be challenging as it mimics many other diseases.

## Case Presentation

### Case history / examination

In January 2019 a 42 year old male presented with fever, extensive widespread skin lesions and painful oral ulcers to the emergency department of Imam Reza hospital of Mashhad city, Iran.

In physical examination he had a fever of 39°C, multiple ulcers on the soft palette and tongue, extensive reddish purple papules and plaques on the face, scalp, torso and limbs. The plaques in some areas were

inflamed and ulcerated with serous discharge and some were erythematous with thick hyperkeratotic crusts. (Figures 1 and 2) Also bilateral orbital edema, cushingoid appearance and two atrophic scars on the right cheek were witnessed.

The lesions had started two years ago and ever since there were periods of remissions and recurrences. During the last few days the patient had experienced deterioration of skin lesions and development of oral ulcers and fever.

There was a history of CL lesions on the face four years ago and he had undergone treatment with intralesional antimoniate which was followed by partial remission.

The patient reported arbitrary use of prednisolone 30mg per day due to Rheumatoid Arthritis (RA) for 10 years.

The patient was febrile and had some punched-out ulcers on his soft palate causing severe dysphagia alongside extensive and widespread skin lesions which in some areas were ulcerated with serous discharge promoting a primary diagnosis of bacteremia status and reactivated HSV due to immunosuppression. Patient received antibiotics and oral acyclovir in the emergency department.

### Differential diagnosis, investigations and treatment

Biopsy was taken from skin lesions with variable differential diagnoses such as psoriasis, sarcoidosis, sweet syndrome and mycobacterial infections and connective tissue diseases. Histopathology showed dense lymphohistiocytic infiltrates in the dermis. The histiocytes had foamy cytoplasm with numerous leishman bodies inside. (Figure 3)

Skin smears from four other lesions from different areas of the body were collected and investigated with Giemsa staining which showed numerous *Leishmania* amastigotes in all four areas. Polymerase Chain Reaction (PCR) test reported *Leishmania. tropica* species.

The patient received intramuscular Meglumine Antimoniate (Glucantime) injections, 20mg/kg/day. Improvement of clinical features was remarkable in ten days. Because of the increase in liver enzyme, the treatment was stopped for few days and restarted with a lower dosage and continued for 28 days.

### Outcome and follow-up

After 28 days treatment with Glucantime clinical improvement was significant (Figures 1 and 2). Patient was referred to rheumatologist and endocrinologist for treatment of RA and corticosteroid dependence. Subsequently, systemic corticosteroid was tapered.

### Discussion

Immunosuppressive treatments alter clinical manifestations of CL and it has been shown in previous studies that immunocompromised patients experience more severe symptoms compared to the immunocompetent. (5) Unconventional manifestations of this disease can cause a delay in diagnosis resulting in harsh circumstances such as scarring, defacement and even disablement. (6) The current case mimicked different conditions. The initial lesions were scaly and erythematous papules and plaques that covered both upper limbs, torso and distal of lower limbs which resembled psoriasis. Plaques in some areas were ulcerated with serous discharge mimicking mycobacterial infection. Some of the lesions were edematous and inflamed which alongside the poor general condition of the patient and fever resembled sweet syndrome. The oral ulcers made the primary health care practitioner think of herpetic infection due to immunosuppression. The association of histopathology with PCR led to diagnosis.

In this case presentation we report a recurrent and disseminated CL with a very rare and unusual clinical presentation. Disseminated Cutaneous Leishmaniasis is a rare manifestation of CL and is linked to the cellular immunity. (4) To the best of our knowledge there have been three reports of disseminated CL after treatment with immunosuppressive drugs following organ transplants. (7,8,9) Two cases of disseminated leishmaniasis by *Leishmania. tropica* were reported in patients with HIV in Iran which presented with multiple skin lesions

on face and extremities. (10) Also Alcover et al reported a case of diffuse CL by *Leishmania.infantum* in a Patient with psoriasis and RA undergoing anti-TNF therapy. (11)

There have been some reports on reactivation of CL infection in patients receiving immunosuppressive treatments for RA, the first case of CL reactivation was witnessed in a RA patient under treatment with systemic corticosteroids in 2005 but it was caused by *Leishmania.donovani* and presented with a single ulcerated lesion. (12) Another report of Visceral and Mucocutaneous Leishmaniasis recurrence was in a Belgian woman with a long history of severe RA who had been treated with etanercept, ciclosporin and methylprednisolone. (13) The current case correlates with earlier reports that CL can cause unconventional clinical manifestation and reactivation in patients receiving immunosuppressive treatments, however the appearance and extent of our patient's lesions were different from any reports that's been done. Also unlike other similar studies the patient was solely being treated with prednisolone.

The standard treatment for disseminated leishmaniasis is systemic antimonial compounds (14, 15), which in this case the aforementioned treatment course was completed and clinical improvement was witnessed.

## Conclusion

Cutaneous Leishmaniasis can cause extraordinary and unconventional clinical manifestations in immunosuppressed patients which could easily be mistaken with other diseases, and diagnosis needs further evaluations and special consideration.

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## Conflict of interests

None declared.

## Authors' contributions:

M.M.; Designed, interpreted the patient data and co-wrote the paper. R.A.; Interpreted the patient data, follow upped the patient and co-wrote the paper. Y.N. and M.T.; Designed and interpreted the patient data. V.M.; Performed the histological examination, Supervised the research and revised the manuscript. All authors read and approved the final manuscript.

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## Figures:





**Figure 1.** Erythematous and indurated lesions with thick hyperkeratotic crusts before treatment (A) and



after 28 days treatment with Glucantime. (B)

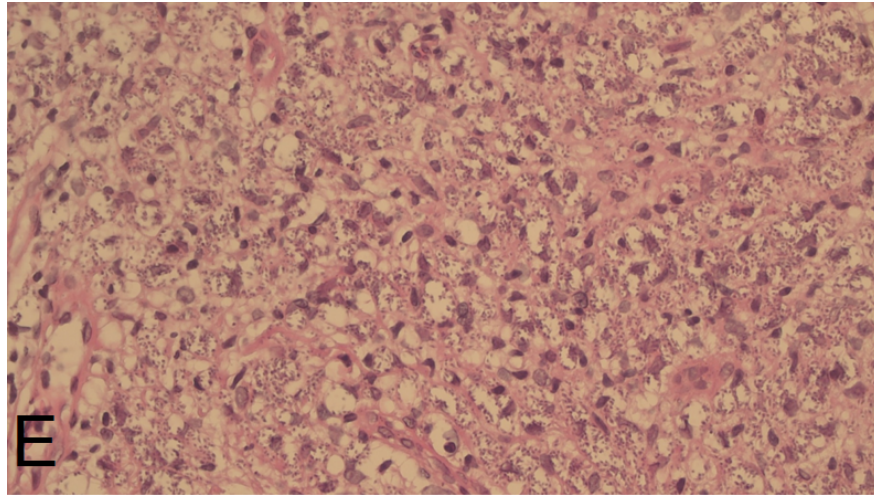




**Figure 2.** Cutaneous lesions on patient's torso before treatment (C) and after 28 days treatment with



Glucantime (D)



**Figure 3.** Hematoxylin and Eosin stain showing dense lymphohistiocytic infiltrates in dermal layer and histiocytes with foamy cytoplasm and numerous leishman bodies.



