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**Case Series**

# Cutaneous leishmaniasis: an enigma of rare clinical presentations

**Rohini Sharma***

Department of Dermatology, GMC Rajouri, Jammu and Kashmir, India

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***Correspondence:**
Dr. Rohini Sharma,
E-mail: dr.rohini_sharma@yahoo.co.in



## ABSTRACT

Leishmaniasis is a neglected tropical disease caused by leishmania parasite. It is divided into two groups: old world species like *L. major, L. infantum*, and *L. tropica* and new world species such as *L. amazonensis, L. mexicana, L. panamensis, L. braziliensis,* and *L. guyanensis.* In India, cutaneous leishmaniasis (CL) cases are mostly seen in Rajasthan, Delhi, Himachal Pradesh, Haryana, Jammu and Kashmir with few isolated reports from south India as well. J and K over the past few years has emerged as a hot spot of CL. Although majority of the cases show classical presentation, atypical presentations mimicking various dermatoses are witnessed. As a clinician, we need to have a high index of suspicion to confirm the diagnoses. Moreover, in an endemic area, it is imperative for the physician to be well acquainted that any atypical lesion, especially chronic form, should be investigated for CL.

**Keywords:** Cutaneous leishmaniasis, Atypical presentation, Biopsy

## INTRODUCTION

Leishmaniasis, recognized as a neglected tropical disease, comprises a group of protozoan diseases, caused by leishmania parasite. Various studies have put the current estimates about incidence of CL at 700,000 to 1.2 million cases per year.[1] Till date, more than 20 species of leishmania parasite have been identified till date; these are divided into two groups: old world species like *L. major, L. infantum*, and *L. tropica* and new world species such as *L. amazonensis, L. mexicana, L. panamensis, L. braziliensis*, and *L. guyanensis*.[2] In India, CL cases are mostly seen in Rajasthan, Delhi, Himachal Pradesh, Haryana, J and K with few isolated reports from south India as well.[3] J and K over the past few years has emerged as a hot spot of CL. Although majority of the cases show classical presentation, atypical presentations mimicking various dermatoses are witnessed. A high index of suspicion is required to confirm the diagnoses.[4] This was a prospective observational case series and in this case series, we described four such case scenarios, who reported to our tertiary institute in the region of Pir Panjal area of Rajouri. A detailed history, clinical examination and biopsy was done. All the cases reported histopathological came out as cutaneous leishmaniasis.

## CASE SERIES

### *Case 1*

A 70 year old female from Poonch area, presented to the skin outpatient department (OPD) with a skin lesion present for the past 4 months.

The lesion, according to the patient started as a small papule later evolving into a plaque over the whole of the lower lip. On examination, whole of the lower lip was hypertrophic with oozing and crusting and at places verrucous appearances. The margins of the plaque had a violaceous hue. A clinical diagnoses of squamous cell carcinoma was made. Biopsy was done and sent to for histopathological examination. The histopathological slides demonstrated Leishman-Donovan (LD) bodies (Figure 1 and 2).

The patient was started on intramuscular sodium stibogluconate 6ml intramuscular in divided doses. The patient was given this for 3 weeks and later shifted to weekly intralesional sodium stibogluconate (SSG) due to non- tolerability for 6 weeks on resolution of the plaque.

**Figure 1: Leishman-Donovan (LD) bodies.**

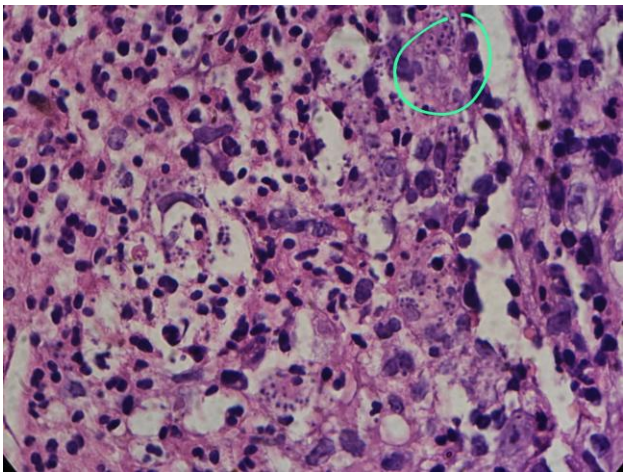

**Figure 2: The histopathological slides demonstrated Leishman-Donovan (LD) bodies**

***Case 2***

A 35-year-old male hailing from a far-flung area of Pir-Panjal region presented to the skin OPD with a skin lesion over the right forearm for the past 7 months. No history of any preceding trauma or any bite was there.

On examination there was a single hypertrophic proliferative lesion with overlying verrucous surface and surrounding erythema. The patient was put for biopsy and differentials of verrucous tuberculosis, chromoblastomycosis, sporotrichosis were kept. The histopathological examination to our surprise demonstrated LD bodies (Figure 3 and 4).

**Figure 3: Surprise demonstrated LD bodies.**

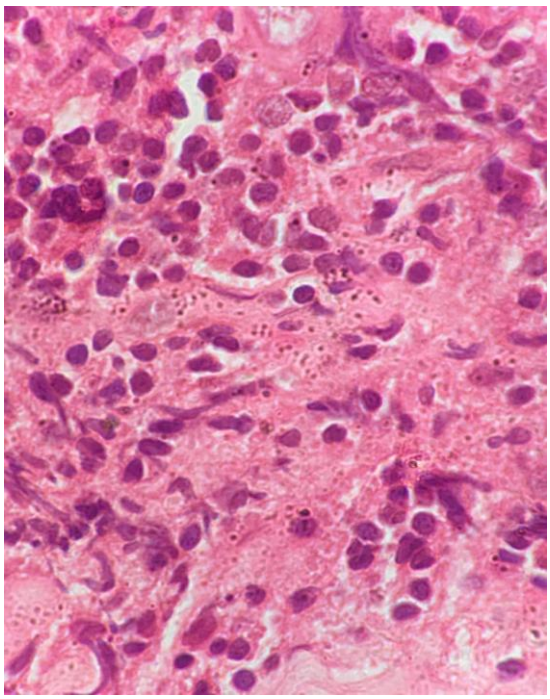

**Figure 4: The histopathological examination to our surprise demonstrated LD bodies.**

The patient was put on intramuscular sodium stibogluconate for 6 weeks leading to resolution of the lesion (Figure 5).

**Figure 5: Intramuscular sodium stibogluconate for 6 weeks leading to resolution of the lesion**

***Case 3***

A 57-year-old female, housewife, presented to our department with a single plaque over the right forearm for the past 11 months.

On examination, there was a single erythematous ill-defined plaque, showing areas of scarring present over the lateral part of right forearm near the wrist (Figure 6). No history of ulceration was present with no family history of similar lesion. There was no history of anti-tubercular treatment taken. Clinically, differentials were kept as chromoblastomycosis, keloid, sporotrichosis and lupus vulgaris were kept. KOH examination was done to rule out any copper penny bodies but was negative. Mantoux test and ESR were negative rest investigations didn't support cutaneous tuberculosis. Culture for sporotrichosis was negative. The histopathological examination showed granulomatous changes in the dermis with multinucleated giant cells. LD bodies were not seen. In lieu of the endemicity of sporotrichosis, a clinical diagnosis of CL was made. The patient was given weekly intralesional sodium stibogluconate for 8 weeks and the lesion was resolved.

**Figure 6: Scarring present over the lateral part of right forearm near the wrist.**

***Case 4***

Here we present a 11-year-old girl, presenting to our OPD with a plaque over the upper lip for 6 months. On examination a single well to ill-defined plaque was present over the upper lip, measuring 8×4 cm. The margins were erythematous with crusting over the surface. On palpation, the plaque was non tender and friable. Crusts were removed; biopsy was taken. The histopathology showed granulomatous inflammation with histiocytes containing the amastigote forms. The patient was started on intralesional sodium stibogluconate weekly but was lost to follow up.

## DISCUSSION

CL is a parasitic disease seen in various countries across the world where it is endemic. In perspective of Jammu and Kashmir, it was first reported from Kashmir valley in 2009 and since then there is no looking back.[5] The twin districts of Rajouri and Poonch have emerged as the hot spots in recent times.[4] the topography and geography of this region do facilitate the transmission of leishmanial infection that include low altitude (<600 m above sea level), heavy annual rainfall, mean humidity above 70%, a temperature range of 15-38°C, abundant vegetation, and alluvial soil.[6]

Here in we have described a series of CL with atypical clinical manifestations who reported to our OPD. All these patients had varied clinical presentations with prolonged durations. Differentials of squamous cell carcinoma, lupus vulgaris, sporotrichoses was also made alongside. Histopathological confirmation of LD bodies was the clue to diagnoses in all. This highlights the requirement of a high index of suspicion in non-classical

cases of CL which has been reported in other studies as well.[7-9]

Various treatment modalities have been used in the management of CL. The most effective being pentavalent antimonial compounds yielding the best results in both adults and children.[10] In our study we gave intramuscular and intralesional according to WHO protocols giving us good clinical response. The finding of clinical response corroborated with decrease in erythema, induration and resolution of the lesion. Various other studies have also found good clinical improvement with intramuscular and intralesional SSG.[11]

## CONCLUSION

The CL with atypical variants and presentations can often be subjected to misdiagnosis or delayed diagnoses further accentuating the patients' morbidity. Thus, we need to keep an iota of doubt in favour of CL in such lesions presenting in area with high endemicity.

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