

A new focus of cutaneous leishmaniasis in Jammu division of Jammu and Kashmir State, India

Neenu Kaul, Vipin Gupta, Subhash Bhardwaj¹, Devraj Dogra, Naina Dogra

Departments of Dermatology, Venereology and Leprology and ¹Pathology, Government Medical College, Jammu, Jammu and Kashmir, India

Address for correspondence:

Dr. Neenu Kaul,
House Number 109, Lane 2,
Roop Nagar Enclave,
Jammu - 180 013,
Jammu and Kashmir, India.
E-mail: shuhulmasti@gmail.com

ABSTRACT

Background: Cutaneous leishmaniasis in India is mainly confined to the deserts of Rajasthan; some cases have been reported from the dry north-western half of the Indo-Gangetic plain, including Punjab, Himachal Pradesh, Delhi and Varanasi. **Aims:** To highlight a new focus of cutaneous leishmaniasis in the Jammu division of Jammu and Kashmir State, previously a non-endemic area. This report presents the clinico-epidemiological and investigative results of 120 new cases of cutaneous leishmaniasis detected between November 2012 and October 2013. **Methods:** The clinical diagnosis of cutaneous leishmaniasis was made using criteria proposed by Bari and Rahman. It was further confirmed by the demonstration of Leishman-Donovan bodies in Leishman stained slit skin smears and skin biopsy specimens, and/or by a satisfactory response to intra-lesional sodium stibogluconate given weekly for 4 weeks. Serial clinical photographs were taken before giving injections and at the end of the 6th week. **Results:** There were 67 females and 53 males with an age range of 8 months to 80 years. The most frequently affected site was the face. Lesions were most commonly of the nodulo-ulcerative type. The number of lesions ranged from 1 to 4. Farmers (28.1%), homemakers (27.2%) and students (27.2%) were significantly over-represented among the occupations ($P < 0.001$). Skin smears and biopsies were positive for Leishman-Donovan bodies in 50.8% and 44.2% cases, respectively. **Conclusions:** There is a new focus of cutaneous leishmaniasis in Jammu division which deserves urgent attention from the public health angle. Further epidemiological studies are warranted to establish the identity of the vector and the strain of *Leishmania* involved.

Key words: Cutaneous leishmaniasis, intralesional sodium stibogluconate, Leishman-Donovan bodies

INTRODUCTION

Cutaneous leishmaniasis continues to be an important public health problem in several parts of the world including India, Pakistan, Afghanistan, Algeria, Iran, Iraq, Peru, Brazil, Africa, Europe, North and South America.^[1] It is transmitted by the bite of an infected female sandfly. Sandflies live in dark, damp places. They acquire the *Leishmania* organisms by feeding on infected animals and humans. Once a sandfly is infected, it can transmit the parasite to both humans and animals.^[2]

A typical lesion of the localized form of cutaneous leishmaniasis is a painless papule that enlarges over a period of several days to weeks to form a nodule or plaque. Ulceration is common. Most people have one or two lesions varying in size from 0.5 to 3 cm in diameter, usually on exposed parts of the body such as the face, arms or legs. The average time from nodule to scar is 2 to 6 months in cutaneous leishmaniasis due to *Leishmania major* and about 6–15 months in *Leishmania tropica*.^[3] Atypical clinical variants of the disease have been

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reported including paronychia, chancreiform, annular, palmoplantar, zosteriform and erysipeloid forms.^[4]

In an endemic area, cutaneous leishmaniasis can be diagnosed by its clinical appearance. Laboratory diagnostic techniques such as slit skin smear, impression smears, culture and skin biopsy are required to facilitate diagnosis when the lesions of cutaneous leishmaniasis appear in a non-endemic area, when superadded infection alters the clinical picture or when any unusual variant is seen.^[1]

Pentavalent antimonials with their consistent efficacy remain the drugs of choice for treating localized cutaneous leishmaniasis. WHO recommends intralesional sodium stibogluconate therapy for early and localized cutaneous leishmaniasis.^[5] This regimen has some side effects such as pain and swelling at the injection site which are relatively minor when compared to serious side effects seen with systemic therapy such as cardiovascular toxicity, neurotoxicity, renal toxicity and bone marrow hyperplasia.

Hitherto, the Jammu region has never been reported to harbour endemic foci, and so far, no study on cutaneous leishmaniasis has been conducted in this region; we are reporting our findings to highlight a new focus of cutaneous leishmaniasis in the state.

METHODS

The present study was a hospital-based, descriptive, clinical study conducted in the department of dermatology in collaboration with the pathology department at Government Medical College, Jammu.

One hundred and twenty consecutive clinically diagnosed cases of cutaneous leishmaniasis constituted the subject material for the study. Pregnant women, children <3 months, patients who had received some definitive treatment for cutaneous leishmaniasis in the past and patients complaining of fever and cough with or without hemoptysis or any other systemic illness were excluded. A detailed history was taken; epidemiological data, onset and duration of skin lesion (s), history of associated itching or pain, history of sleeping outdoors, insect bite, house design and history of a similar illness in the family or neighborhood were recorded on printed proformas. After obtaining informed consent, every patient was subjected to general physical, systemic, cutaneous and mucosal examination.

The clinical diagnosis of cutaneous leishmaniasis was made using the clinical criteria proposed by Bari and Rahman and this was further confirmed, whenever possible, by the demonstration of Leishman-Donovan bodies in Leishman stained slit skin smears and hematoxylin and eosin stained skin biopsy sections.^[1]

Cytological examination Skin biopsy Sample collection was undertaken by a dermatologist. The edge of the lesion was squeezed between the thumb and forefinger to make the area bloodless. A 5 mm long and 2 mm deep incision was made in pinched skin with a small-bladed scalpel. It was rotated at right angles to the cut and the incision was scraped several times in the same direction thus collecting tissue fluid and pulp on one side of the blade. This was gently smeared on at least two glass slides, immediately fixed in 95% ethanol and stained with Leishman's stain. At least 100 different well-stained and preserved fields, mainly in and around the vicinity of mononuclear cells, were examined under the supervision of a senior pathologist at Government Medical College, Jammu, using light microscopy, first under high power and then with the oil immersion lens to identify Leishman-Donovan bodies, both intracellular within macrophages or lying extracellularly.

A skin biopsy was taken from each patient. Hematoxylin and eosin stained sections were examined under the supervision of a senior pathologist at Government Medical College, Jammu.

Therapeutic trial

Parasite-negative but clinically suspected cases of cutaneous leishmaniasis were given a therapeutic trial of intra-lesional sodium stibogluconate (100 mg/ml) weekly, for 4 weeks. The volume injected into each lesion was 0.5–5 ml per injection, depending on the size of the lesion. The response was noted at the end of 6th week. Clinical parameters for assessing response were disappearance of erythema and reduction in lesion size and induration. Serial clinical photographs were taken to aid in response assessment. Those cases showing satisfactory to excellent response to the therapeutic trial were also considered to be confirmed cases of cutaneous leishmaniasis, and were included in the study. Culture and polymerase chain reaction could not be done due to non-availability of these techniques in our hospital.

Statistical analysis of the data was performed using Epi Info (version 6), downloaded from the website of CDC

Atlanta, free of cost. Chi-square test was applied to the data depicting the most frequently appearing clinical finding among the groups; $P < 0.05$ was considered significant. The study was reviewed and approved by the institutional ethics committee.

RESULTS

A total of 120 cases of cutaneous leishmaniasis were included in the study. There were 145 lesions (range 1–4 per patient) observed in 120 patients. Patient age ranged from 8 months to 80 years (mean age, 29.05 years). The largest number of patients was seen in the age group of 21–40 years. A slight female predominance was seen; 67 (55.8%) were females and 53 (44.2%) were males. Among the various occupations, farmers (28.1%), homemakers (27.2%) and students (27.2%) were significantly over-represented ($P < 0.001$).

Nodulo-ulcerative lesions were predominant [Figure 1], seen in a total of 42 (35%) patients, followed by erythematous, edematous plaques in 39 (32.5%) [Figure 2], nodules in 34 (28.3%), papules in 4 (3.3%), and sporotrichoid pattern in one patient.

The number of lesions ranged from 1 to 4. A single lesion was present in 101 (84%) cases, whereas multiple lesions were seen in 19 (16%) cases.

The face [Figure 3] was the most commonly affected site followed by the neck, upper limbs, lower limbs and upper trunk.

The duration of skin lesions before presentation to us varied from 1 month to 12 months. However, 71 (59.2%) of our patients presented with skin lesions of 3–6 months' duration. Most of our patients lived in

kaccha houses and a history of sleeping outdoors was noted in 44 (36.7%) patients.

Leishman-Donovan bodies were demonstrated in 63 (52.5%) cases. Skin smear was positive in 61 (50.8%) patients [Figure 4]. Biopsy material also demonstrated Leishman-Donovan bodies either within macrophages and or extracellularly in 53 (44.2%) patients. In 10 (8.3%) patients, we were able to visualize parasites only on slit skin smear but skin biopsy did not show any Leishman-Donovan bodies. Likewise, in 2 (1.7%) patients, we were able to demonstrate parasites only on histology.

Fifty seven patients did not show any Leishman-Donovan bodies either on skin smear or histology but responded well to a therapeutic trial of intra-lesional sodium stibogluconate given weekly for 4 weeks. The clinical diagnosis was confirmed by a positive response to therapeutic trial in all these 57 patients and, thus, they were also included in our study as cases of cutaneous leishmaniasis.

The other associated histopathological findings were epidermal and dermal changes. Epidermal changes were seen in 88 (73.3%) cases. Common changes included ulceration (44, 36.7%), acanthosis (34, 28.3%) and hyperkeratosis (32, 26.7%). Rare findings included parakeratosis in three patients, follicular plugging in two and epidermal atrophy in one patient. In the dermis, well-formed epithelioid cell granulomas without necrosis were seen in 60 (50%) cases and epithelioid cell clusters in 33 (27.5%) cases. Lymphocytes were seen in all 120 (100%) patients. Plasma cells were demonstrated in 97 (80.8%) cases, histiocytes in 75 (62.5%) cases, eosinophilic leukocytes in 49 (40.8%) cases, Langhans giant cells in 36 (30%) cases and neutrophils in 5 (4.2%) cases [Figure 5].

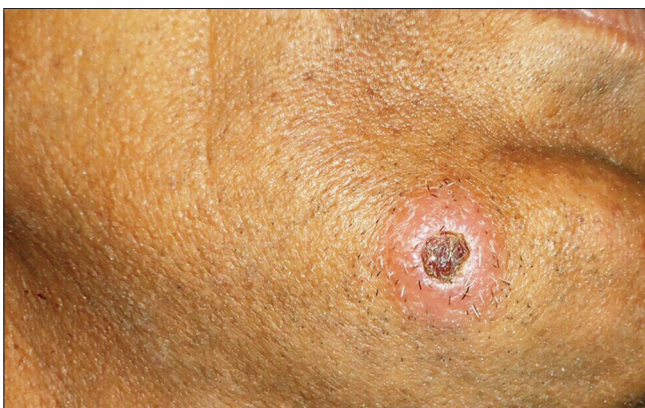


Figure 1: Well-defined erythematous nodular lesion with central ulceration and crusting



Figure 2: Well-defined erythematous, edematous crusted plaque present over bridge of nose and spreading to involve medial area of both cheeks



Figure 3: Single well demarcated ulcerated plaque with hemorrhagic crusting present over lower lip

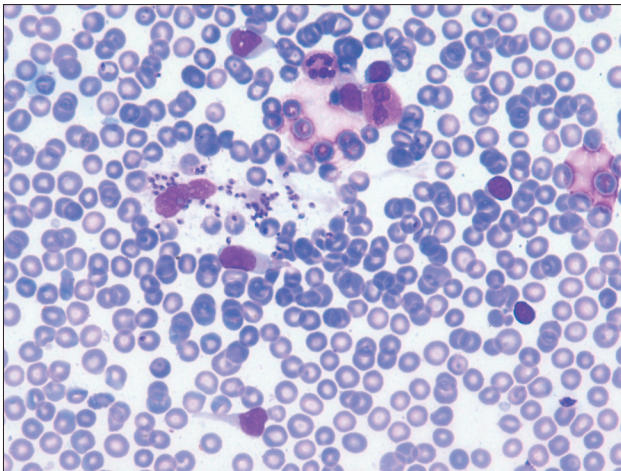


Figure 4: Slit skin smear (Leishman, x400) showing numerous extracellular Leishman Donovan bodies lying free in the vicinity of macrophages present in the centre of smear

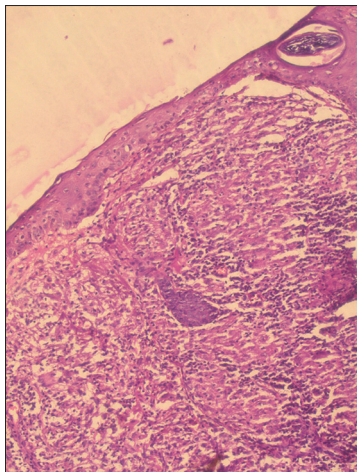


Figure 5: Dermis showing numerous well formed epithelioid cell granulomas along with heavy chronic lymphomononuclear and plasmacytic infiltrate (H and E, x400)

DISCUSSION

Cutaneous leishmaniasis, once restricted to certain areas, is now spreading to places that were previously known to be non-endemic and new foci of infection

are regularly being encountered in India as well as Pakistan.^[2-4] Cutaneous leishmaniasis is a vector-borne protozoal infection of the skin caused by several species of *Leishmania*, mainly *Leishmania major*, *Leishmania tropica* and *Leishmania aethiopica* in the Old World, and *Leishmania braziliensis* and *Leishmania mexicana* in the New World. The usual clinical picture of cutaneous leishmaniasis varies from erythematous papules to nodulo-ulcerative forms, and mostly, lesions are seen on the exposed parts of the body.^[6] In an endemic area, cutaneous leishmaniasis can be diagnosed by its clinical appearance. Laboratory diagnostic techniques such as slit skin smear, impression smears, culture and skin biopsy are required to facilitate diagnosis when the lesions of cutaneous leishmaniasis appear in a non-endemic area, when superadded infection alters the clinical picture or when any unusual variant is seen.^[1] Demonstration of amastigotes in skin smears or skin biopsy and culture are confirmatory. Modern methods for the diagnosis of leishmaniasis are immunofluorescence, use of monoclonal antibodies, DNA probes, polymerase chain reaction and electron microscopic studies.

All our patients belonged to three districts of Jammu division; Doda, Kishtwar and Poonch. These areas lie in hilly terrain in the middle and outer Himalayan range; they experience hot summers and also cold winters, when temperatures may fall to 0°C or lower (temperate climate). Rainfall is minimal throughout the year. The topography of these areas favors the growth of sandflies. Over the past few years, there has been an increase in the migrant population in these areas hailing mostly from Himachal Pradesh and Pakistan, which are known endemic areas for leishmaniasis. However, none of our patients had visited areas known to be endemic for cutaneous leishmaniasis during the previous one year.

We observed younger patients to be frequently affected by the disease. A little over 40% of our patients were between 21 and 40 years old. One explanation could be that people of this age have more outdoor activities than other age groups and hence are exposed more to the bites of sandflies. These findings are consistent with the earlier studies by Simi *et al.*, Sharma *et al.* and Sharma *et al.*, where a high prevalence of cutaneous leishmaniasis in young patients was reported.^[7-9]

There was a female preponderance in our study as 67 (55.8%) out of the 120 patients were women. This

may be due to greater cosmetic concerns in women. Sharma *et al.* and Sharma *et al.* have also observed a slightly higher female preponderance in their respective studies.^[8,9] However, Aara *et al.* described male preponderance in their study which they justified on the basis that men wore less clothing and worked more in open environments than women.^[10]

Among the various occupations, farmers, homemaker and students were significantly over-represented in this study. Our results are comparable with the study conducted by Abdellatif *et al.*, who also observed that farm workers, homemakers and students were at a high risk of contracting cutaneous leishmaniasis.^[11]

Nodulo-ulcerative lesion was the most common type of cutaneous leishmaniasis in this study. Similar findings were reported by Sharma *et al.*^[8] The atypical sporotrichoid variant was seen in one patient; the same variant has also been observed by Raja *et al.*, Markle and Makhoul, and Karıncaoglu *et al.*^[3,12,13] The sporotrichoid pattern is thought to represent an immune reaction from direct lymphatic extension of *Leishmania* organisms or antigens.^[4] The diverse clinical spectrum of cutaneous leishmaniasis results from the complex interaction of a number of factors such as the type and duration of clinical lesions, strain of organism, geographic location, parasitic load, disease reservoir and host immunocompetence.^[14]

A single lesion was found in 101 (84%) of our patients and 2–4 lesions were seen in the remaining 19 (16%). These findings are consistent with the earlier studies by Sharma *et al.*, Sharma *et al.*, Aytakin *et al.* and Hepburn *et al.*, who found single lesions in 74%, 65%, 47% and 71% patients respectively.^[8,9,15,16]

In our study, face (87.6%) was the most commonly affected site followed by the neck (4.8%), upper limbs (4.2%), lower limbs (2.1%) and upper trunk (1.4%). This observation conforms to the well-established clinical picture of cutaneous leishmaniasis. Sharma *et al.* also noted that lesions were mostly on the face in their patients.^[8]

A history of sleeping outdoors was seen in 37% of cases. Sleeping outdoors or indoors without a bed net increases the risk of sand fly bites since these insects are most active in the evening and at night. This finding is consistent with that of Al-Taqi and Behbehani.^[17] In our study, the majority of patients (65%) lived in

kaccha houses. Masood *et al.* had reported that all his 40 patients lived in kaccha (mud) houses.^[18] Crevices in kaccha houses are one of the preferred breeding sites of sandflies.

Skin smears demonstrated amastigotes (Leishman-Donovan bodies) in 50.8% of our cases. The rate of skin smear positivity has varied in different studies. Simi *et al.* reported a smear positivity rate of 57.2%,^[7] while the figure reported by Sharma *et al.* was only 37%.^[9]

Amastigotes were visualized in skin biopsies in 44.2% of our patients; a comparable figure of 43.8% was reported by Rawlins *et al.*^[19] However, Kubba *et al.* demonstrated parasites in 70% of tissue biopsies.^[20] We found that Leishman-Donovan bodies could be detected (both on skin smear and biopsy) in 61.7% of patients whose lesions had appeared within the previous 6 months but this figure dropped to 33.3% for patients whose lesions were 6–12 months old. The success of demonstration of the organism depends on a multitude of factors including the duration of the lesion. The detection of the parasite becomes more difficult as infection progresses.^[21]

In 10 patients, we were able to visualize parasites only on slit skin smear but skin biopsy did not show any Leishman-Donovan bodies. Failure to demonstrate parasites on histology could be explained by the Bahamdan *et al.* postulation that *Leishmania* organisms appear larger in skin smears and are smaller and scantier on histopathology due to repeated processing of biopsy specimens with dehydrating solutions.^[22]

There was no difference in type or size of skin lesions in patients with Leishman-Donovan body positivity as compared with Leishman-Donovan body negative cases.

CONCLUSION

The present study has highlighted a new focus of cutaneous leishmaniasis in Jammu division of Jammu and Kashmir State which poses a considerable public health hazard. Further, epidemiological studies are warranted to establish the exact identity of the vector and the strain of *Leishmania* involved.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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