Case Report

# Cold cellulitis

## *An unusual presentation of cutaneous leishmaniasis*

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## ABSTRACT

We describe a case of cutaneous leishmaniasis, which presented as what we call "cold cellulitis". This may be differentiated from classical cellulitis/erysipelas by 1. Lesser local reaction like pain and tenderness 2. No systemic symptoms 3. No leucocytosis 4. Negative bacteriological and serological assay for bacterial cellulitis/erysipelas, 5. Longer course of illness and 6. Failure to respond to antibiotics. However, awareness of this type and frequent skin smears or skin biopsy will settle the right diagnosis. A review of different leishmanial presentations is given for comparison.

**Keywords:** Cold cellulitis, cutaneous leishmaniasis, leishmanial cold cellulitis

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It is not unusual to find an indurated erythematous margin around a leishmanial lesion. It is usually part of it. To have such a marked erythematous indurated swelling so much so that it obscures partially the features of leishmaniasis, is an abnormal phenomenon. This assumes importance as it may deviate the diagnosis towards bacterial cellulitis. Here we are reporting an intriguing case of cutaneous leishmaniasis (CL) of this type, presenting as cellulitis like lesion or what we here describe as "leishmanial cold cellulitis".

**Case Report.** A sixty-year-old Yemeni woman was referred to us for sudden appearance of a skin lesion on the face since 7 days. She suspected it to be an insect bite near the right eye. She had a hot heavy feeling at the site and was anxious for it. On examination **(Figure 1)** the patient was not febrile. There was diffuse ill-defined indurated erythematous surface on the central region of the face "nose, cheeks and upper lips" with puffiness. Orange peel sign was visible on it. The lesion was angry looking, but surprisingly it was not tender. Two central hyperkeratotic ulcerations super mounted it. Submandibular lymph node was palpable. Systemic examination was otherwise normal. Blood picture was within normal limits. White blood cells count was 6,600 with 68% segmented cells, 24% lymphocytes, 6% eosinophils and 2% monocytes. anti-streptolysin O titer was normal. Specimen from the ulcers for Gram stain and culture were negative. Swabs for culture from the nose, throat and the right eye were negative as well. Patient was admitted in the surgical ward and was treated as a case of cellulitis with penicillin intravenous. But she did not improve. Smears for Leishmania Donovan bodies (LD bodies) were negative "possibly due to the severe inflammatory reaction". Later they did appear

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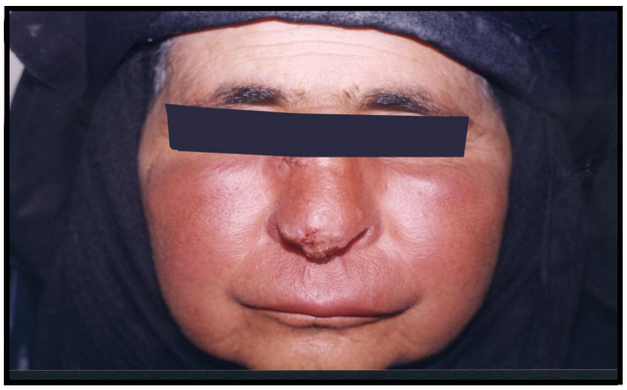

**Figure 1 -** Cutaneous leishmaniasis of the cold cellulitis/erysipelas type photo taken after 3 weeks of illness.

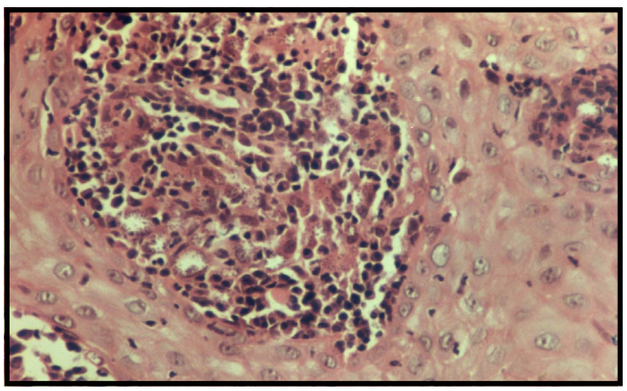

**Figure 2 -** A microscopic field: Leishmania Donovan bodies within the macrophages.

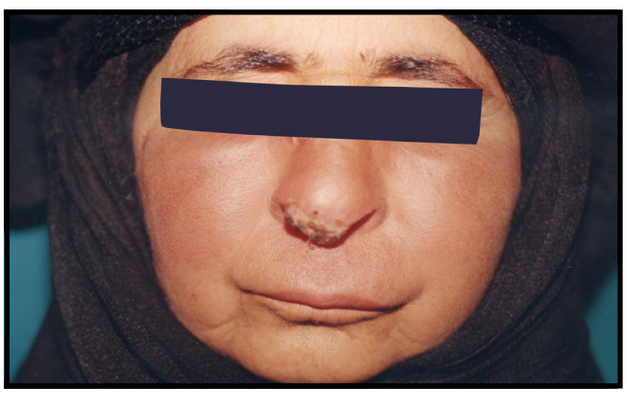

**Figure 3 -** The patient after 4 weeks of antimonial therapy.

on skin biopsy sections taken from the ulcer on the nose (**Figure 2**). The histopathological picture was suggestive of leishmaniasis as well. A course of pentavalent antimony for 4 weeks was given with marked improvement (**Figure 3**).

**Discussion.** The first look at the lesion especially on the upper lip suggests the diagnosis of cellulitis/erysipelas (C/E). It shares C/E in having erythema of acute onset, swelling, heat and lymphadenopathy. The 2 ulcers on the nose might suggest the pre-existence of blisters which may appear in C/E. Or else they might suggest 2 sites of an unknown insect bites which might have acted as a possible portal of the C/E infection. This is why the case was provisionally treated as C/E infection in the beginning. However, it differs from classical C/E lesion in: 1. The absence of pain and tenderness despite the severe inflammatory reaction noted. 2. No constitutional symptoms like fever, malaise etc, 3. Hematological picture did not show any leucocytosis or shift to the left as almost always occurs in C/E, 4. The patient failed to respond to antibiotics, 5. The disease had a longer and calmer course than that of classical C/E. These differences made us think of some other diagnosis. As the erosions on the nose gradually became hyperkeratotic (**Figure 1**) the possibility of CL was considered and later confirmed as mentioned above. Although some clinical presentations are indicative of the causative leishmanial parasite, it is not always a correct correlation.[1] There is a big variety in the host response so that the lesions are not always characteristic of the causative species.[2] A review of the various presentations of the old world leishmaniasis shows that CL may present in numerous types which include: a) the symptomatic type, where sub-clinical infection which pass unnoticed acts as a natural vaccine.[3] b) the wet 'rural type' due to L. major which presents usually as furunculoid lesions "often in clusters" that crust and ulcerate.[3] This type may be associated with lymphadenitis and secondary satellite nodules around it. The presentation may also be as c) an iceberg nodule where the majority of the lesion dive subcutaneously and shows only the top, d) sporotrichoidal nodules due to lymphatic spread, or d) elephantiasis of the lower limbs due to leishmanial lymphangitis. f) The volcano sore is the most characteristic one,[4] g) The dry urban type due to L. minor presents usually as a few brownish nodules which evolve to plaques and ulcerate.[4] h) The lesion on the face of an elderly individual of white complexion may resemble basal cell carcinoma[5] i) A solitary mucosal lesion due to L. infantum has occasionally been reported,[6] j) Chronic leishmaniasis "leishmania recidivans" usually presents as apple-jelly nodules, that is the lupoid type. Rarely k)

keloidal, l) verrucous, on the lower limbs, and m) psoriasiform types have been described.[7] In Indians, rather than the classical visceral type, n) the diffuse generalized blackness "kala-azar" may develop.[8] However, o) some patients may present directly with post kala-azar dermal leishmaniasis without previous history of visceral involvement.[9] In Africans, CL due to L. aethiopica may present as p) a solitary inflamed lesion on the face and it is seldom severe, especially near the mucosal margin of nose and mouth.[10] It may also present in anergic patients as q) diffuse lepromatoid leishmaniasis. African visceral leishmaniasis due to L. donovani or L. infantum, may have a skin presentation as well. In a few cases, it may present as r) a primary cutaneous leishmanial lesion and rarely w) may be accompanied by mucosal lesions.[8] In the endemic areas of both acquired immunodeficiency syndrome (AIDS) and CL, AIDS may blur the clinical presentation of CL, may occur primary or secondary to AIDS and may either present as a visceral disease, or it may be silent, where the parasites are found accidentally in the biopsy sections along with histopathological findings of Kaposi sarcoma.[11] This round in the diverse leishmanial presentations shows that a C/E type is not clearly included among them, though anecdotal reports of such presentation may be found in literature.[1] This case from Najran, Kingdom of Saudi Arabia, which we are reporting is an example of it. It occurs whenever the inflammatory reaction caused by the leishmania parasite is huge and marked enough to obscure the original leishmanial features and mask partially the correct diagnosis. It was more obvious in the early stage of this case. The lesser pain and tenderness on the leishmanial lesion compared with the classical bacterial cellulitis may be attributed to the local nerve involvement in CL, which diminishes sensation.[4]

In conclusion, CL may present as cellulitis of cold type. The marginal inflammatory reaction which usually accompanies CL becomes severe enough to mask it and mimic C/E. It then may be differentiated from the classical C/E by the cold features highlighted in the text. It resembles the lepromatous cold cellulitis which we described before.[12] Frequent skin smears or skin biopsy will ultimately settle the issue. To know how prevalent this C/E presentation is, reports from other regions/countries are anticipated. We would like to draw the attention to this exaggerated presentation of CL, which we label as cold cellulitis or leishmanial cold cellulitis. This will help diagnose these cases early so that patient will not be mistreated.

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