

Case Report

Cutaneous Leishmaniasis Presenting as a Submandibular Nodule – A Case Report

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Abstract

A 42 year old male from Kinnaur district of Himachal Pradesh presented with a subcutaneous nodule in the submandibular region. It was 2x3 cm in size, firm, mobile and non-tender. Lymphadenitis and sialadenitis were kept as differential diagnosis. Fine-needle aspiration revealed Leishman Donovan (L-D) bodies along with non-caseating epithelioid cell granulomas. We are presenting this report for its rarity.

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Key Words : Cutaneous leishmaniasis, subcutaneous nodule, FNAC.

Introduction

Localized cutaneous leishmaniasis (LCL) in India is due mostly to *Leishmania tropica*. It is mainly endemic in deserts of Rajasthan. Recently, Kinnaur district in Himachal Pradesh has been identified as a new endemic focus for the disease. Though rarely reported as a cause of LCL, *Leishmania donovani* is the predominant pathogen here along with *L. tropica*.¹ We are presenting a rare case from Himachal Pradesh where cutaneous leishmaniasis presented as a subcutaneous nodule in submandibular region without any associated primary lesion.

Case Report

A 42 year old male, a teacher by profession presented with a submandibular nodule to ENT Department. The clinicians kept a differential diagnosis of lymphadenitis and sialadenitis and referred him to the Department of pathology for fine needle aspiration cytology (FNAC).

On examination, the subcutaneous nodule was located on the right side in submandibular region. It was 2x3 cm in size, firm, mobile and non-tender. Overlying skin was inflamed and indurated; there was no ulcer or crust. Fine needle aspiration was done. Giemsa staining showed scattered extracellular and intracellular Leishman Donovan (LD) bodies along with non-caseating epithelioid cell granulomas and groups of histiocytes. The background revealed scattered RBCs. No giant cells, lymphoglandular bodies or salivary gland structures were seen (Figs. 1 & 2). On Ziehl-Neelsen staining no acid fast bacilli were seen. A diagnosis of granulomatous inflammation with presence of L.D. bodies (subcutaneous tissue)

was made. The patient was given intralesional stibogluconate to which he responded well.

On eliciting the history the patient was serving in Kinnaur district for the last three years. He had noticed this nodule about two months back and he was given antibiotics for suspected folliculitis in district hospital. There was no response to treatment.

Discussion

Localized cutaneous leishmaniasis (LCL) is a vector-borne disease commonly caused by the flagellate parasites *Leishmania tropica* or *L. major* in the old world. It is an obligate intracellular parasite. The

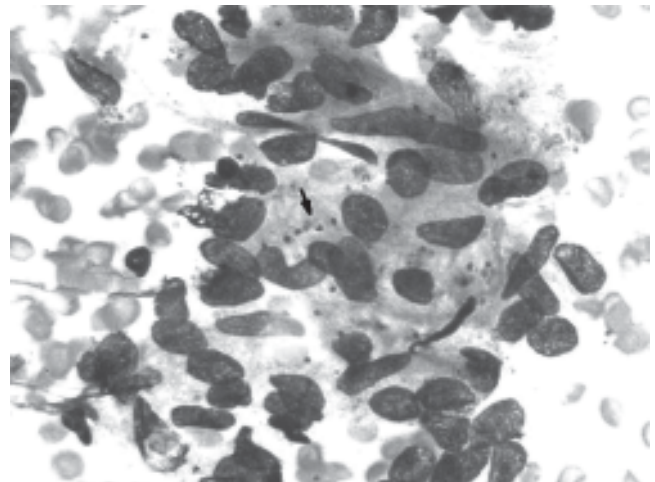


Fig. 1 : Smears show non caseating epithelioid cell granuloma with LD bodies (Giemsa stain, x1000).

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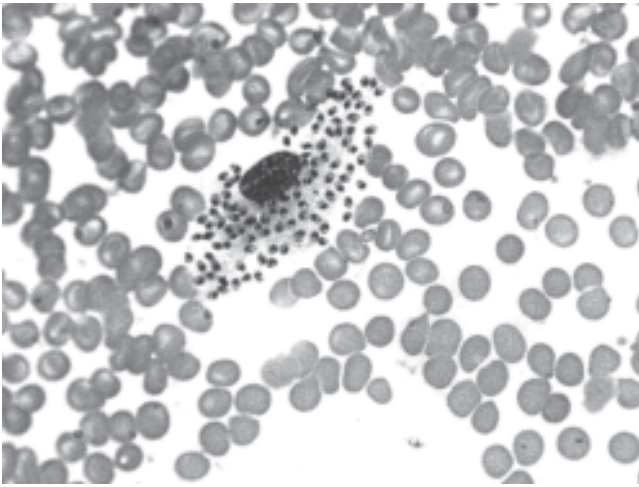


Fig. 2 : Smears revealing intracellular LD bodies (Giemsa stain, x1000).

geographic range of this zoonotic disease is limited by a sandfly vector belonging to genus *phlebotomus* or *lutzomyia*.¹

Cutaneous leishmaniasis (oriental sore) is characterized by slowly evolving, inflammatory lesions that are nodular, nodulo-ulcerative or ulcerative, healing spontaneously with scars in three to twelve months. Acquired immunity is life long. A comprehensive study done by Kubba et al² in Saudi Arabia on cutaneous leishmaniasis due to *L.major* found seven clinical features of diagnostic value: exposed site location (84%), pairing or clustering of lesions (61.72%), skin crease orientation (35.37%), volcanic nodules (30.32%), satellite papules (19.3%), subcutaneous nodules (11.37%) and iceberg nodules (4.63%).²

Occasionally old world cutaneous leishmaniasis can disseminate from primary lesion to subcutaneous tissue and lymph node via proximal lymphatics.^{3,4} The primary lesion is sometimes inconspicuous. The subcutaneous nodule and lymphadenopathy may persist long after the primary lesion has healed.⁴ "Sporotrichoid" lymphangitis frequently accompanies cutaneous leishmaniasis of the new world caused by members of the *L. braziliensis* complex.⁵

In Kinnaur district of Himachal Pradesh, in last few years, the number of new cases of LCL has been increasing reaching almost epidemic proportions. The risk of acquiring LCL increases considerably with developmental projects that have an impact on the environment. In Kinnaur, hydroelectric power projects are being developed over river Satluj.

In this region, 161 cases of LCL were studied by Sharma et al.¹ Most cases were seen from April to September. The lesions mainly involved exposed body parts, being present for 1-6 months. They were usually one to eight in number. Face and neck were affected in all cases. Also all cases in this study presented with nodulo-ulcerative lesions. The rate of smear positivity was 37% and was higher in recent lesions (duration of 1-6 months).

Histopathology analysis in most cases showed chronic granulomatous inflammation and was not diagnostic. *Leishmania* was cultivated and identified by PCR- RFLP. Though rarely reported as a cause of LCL, *L.donovani* is the predominant pathogen in this new focus.¹

Our case acquired infection in Kinnaur, in the month of September with single lesion on exposed part with skin crease orientation. Smear was positive for LD bodies. All these findings helped in reaching the diagnosis of cutaneous leishmaniasis. Though unusual, presentation as a subcutaneous nodule without primary lesion over submandibular region can be kept in the differential diagnosis in endemic areas.

Chemotherapy is justified only with multiple or disfiguring lesions. Intralesional sodium stibogluconate is effective without any major side effects, as was seen in our case.^{1,6}

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