Case Report

Unusual presentation of mucocutaneous leishmaniasis in HIV-infected patient

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Abstract

Leishmaniasis is caused by protozoan parasite of genus leishmania. Visceral leishmaniasis, diffuse cutaneous leishmaniasis, and atypical forms of cutaneous leishmaniasis are common in HIV-infected patients. Our patient presented with an obstructive mass in nasal cavity and was diagnosed as a case of mucocutaneous leishmaniasis. Spontaneous healing of lesions in HIV-infected patients is rare rather they are unresponsive to treatment and have frequent relapses, especially in patients with low CD4 count. However, in our patient, the lesion improved significantly after 2 months of highly active antiretroviral therapy and co-trimoxazole prophylaxis.

Key words: Coinfection, HIV infection, leishmaniasis

INTRODUCTION

Leishmaniasis is a tropical disease caused by protozoa belonging to the group of *Leishmania* species. Few districts of Himachal Pradesh along the Satluj river are endemic for cutaneous leishmaniasis.[1] Leishmaniasis is usually classified as localized cutaneous, mucocutaneous, diffuse cutaneous, and visceral leishmaniasis.[2] The lesion begins as erythematous papule which enlarges to form nodule which then undergo ulceration and crusting. Satellite lesions can also be present. Lesions usually develop over the exposed part of the body such as face, nose, neck, and arms.[2] Lesions heal in a period of 6 months to 1 year. Visceral leishmaniasis, diffuse cutaneous leishmaniasis, and atypical forms of cutaneous leishmaniasis are common in HIV-infected patients.[2] Here, we report a case of mucocutaneous leishmaniasis in an HIV-infected patient who presented with an obstructive mass in the nose and responded well to highly active antiretroviral therapy (HAART) and co-trimoxazole prophylaxis.

CASE REPORT

A 36-year-old male presented with a history of mass left nasal cavity for last 2.5 months. Lesion initially started as red, raised painful lesion which progressively increased in size to involve the whole of the left nasal cavity, anterior nares, and upper lip. The lesion subsequently ulcerated. There was a history of nasal obstruction with difficulty in breathing. There was a history of significant weight loss. There was no history of chronic diarrhea and fever. Patient gave a history of antituberculous drug therapy for cervical lymphadenitis 4 years back. His general physical examination and systemic examination were unremarkable. On mucocutaneous

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examination, there was a single well-defined ulcerated plaque of size 5 cm × 3 cm approximately present inside the left nasal cavity extending to involve tip of nose and upper lip with surface showing erosion and serosanguinous crust [Figure 1]. Examination of oral and genital mucosa showed oral thrush and genital warts. Cutaneous, hair, and nail examination was within normal limit. Possibility of deep fungal infection and lupus vulgaris was kept. Complete hemogram, biochemistry, and chest X-ray were normal. X-ray paranasal sinuses showed opacification in left maxillary sinus with hypertrophy of turbinates. He was positive for HIV test with CD4 count of 19 cells/cumm. There was a history of extramarital sexual contact. His spouse was HIV negative and repeat HIV test at 3 months was also negative. Imprint smear revealed macrophages, sparsely scattered lymphocytes, and intra and extra-cellular Leishman–Donovan bodies [Figure 2]. Skin biopsy showed inflammatory infiltrate in interstitial pattern comprising lymphocytes, few histiocytes with abundant intra- and extra-cellular Leishman–Donovan bodies suggestive of cutaneous leishmaniasis [Figure 3]. On reviewing the history, patient gave history of visit to an endemic area for leishmaniasis. To rule out visceral leishmaniasis bone marrow aspiration and ultrasound abdomen were done which were found to be normal. There was no history suggestive of Kala-azar in the past. Tissue for fungal and acid-fast *Bacilli* culture was negative. Patient was started on antiretroviral therapy (zidovudine, lamivudine, and nevirapine), co-trimoxazole prophylaxis, and oral fluconazole. There was a significant improvement in lesion at 2 months of follow-up [Figure 4].

DISCUSSION

Centers for Disease Control and Prevention has classified leishmaniasis as a neglected tropical disease. Besides, the classic morphology, there can be

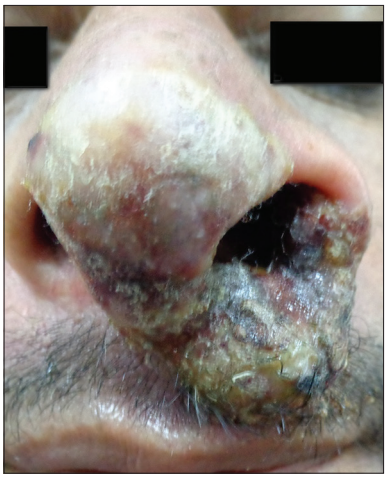

Figure 1: Ulcerated plaque involving left nasal cavity and nose

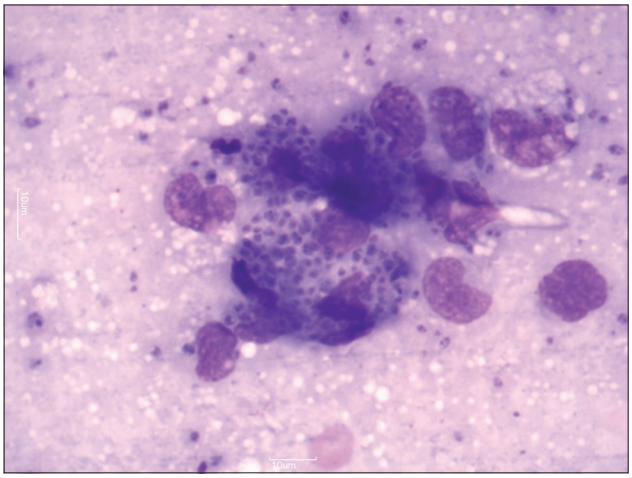

Figure 2: Imprint smear showing intra- and extra-cellular Leishman-Donovan bodies. (Gieimsa stain, ×100)

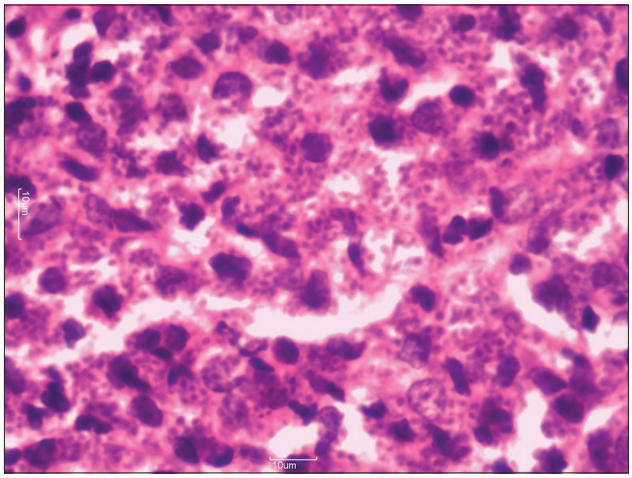

Figure 3: Photomicrograph showing inflammatory infilterate with abundant intra- and extra-cellular Leishman-Donovan bodies. (H and E, ×40)

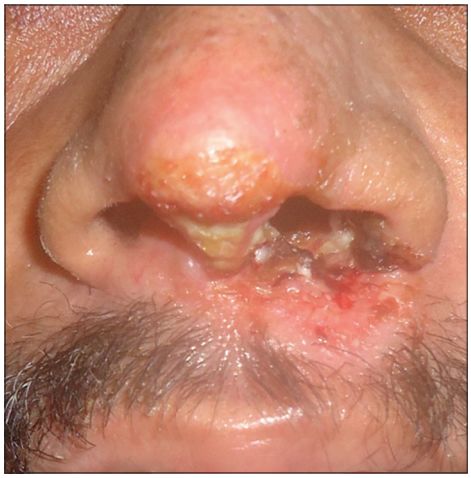

Figure 4: Significant reduction in the size of lesion after 2 months of highly active antiretroviral therapy and co-trimoxazole prophylaxis

atypical presentations such as lesions along Langer's lines, eczematoid, warty, zosteriform, erysipeloid, and sporotrichoid.[2] There may be associated regional lymphadenopathy.[2] Intravenous drug users are at risk of coinfection, especially in visceral leishmaniasis as the contaminated syringes help in the spread of amastigotes.[2] In India, the first case of HIV and *Leishmania* coinfection was reported in 1999 from the sub-Himalayan region.[3] Diffuse cutaneous leishmaniasis is common in HIV-affected patients.[4] Visceral leishmaniasis can occur as an opportunistic infection in late stages of HIV infection. About 1700 cases have been reported worldwide in 33 countries, India having the highest number of cases.[5] Patients with coinfection may have cryptic form of infection. A study revealed that 11% of patients with HIV in Mediterranean Basin had *Leishmania infantum* in the bone marrow aspirates.[4] Cases of leishmaniasis have been reported within lesions of Kaposi sarcoma, squamous cell carcinoma, and basal cell carcinoma in HIV-infected patients.[6] Coinfection leads to increased treatment failure, relapse, drug toxicity, and mortality.[7] In HIV-infected patients, cutaneous dissemination is common in visceral leishmaniasis.[2] Furthermore, atypical lesions and relapses of the disease are common in these patients. Impaired immunity in HIV infection causes the reactivation of latent *Leishmania* infection, whereas leishmaniasis can also promote HIV virus replication and increases progression to AIDS.[8] Mucocutaneous involvement is not a common presentation in old world leishmaniasis, especially nasal involvement. Mucocutaneous leishmaniasis is usually caused by new world species of *Leishmania*, but it can also be caused by *Leishmania aethiopica* and rarely other *Leishmania* species such as *Leishmania tropica*.[1,2] Mucosal involvement may also occur through blood vessels, lymphatics, and contiguous spread.[1] Such dissemination is common in HIV-infected patients.[9] Our patient presented with mucocutaneous leishmaniasis obstructing the nasal cavity with the involvement of maxillary sinuses. We could not identify the *Leishmania* species because of nonavailability of polymerase chain reaction. HIV-infected patients can present with cutaneous lesions similar to immunocompetent patients, but in these patients, visceral leishmaniasis should be ruled out.[2]

The *Leishmania* amastigotes are usually found within the macrophages in immunocompetent patients, but they are present both inside macrophages and free in dermis and subcutaneous tissue in HIV-infected patients with scarce lymphomononuclear infiltrate.[2] In addition, the number of amastigotes is much higher in these patients. Similar findings were observed in our patient also who had a very low CD4 count. Cutaneous leishmaniasis in initial stages of HIV infection responds nicely with intralesional antimony.[2] Purohit *et al*. also reported a case of diffuse cutaneous leishmaniasis in HIV-infected patient who had visceral involvement with bone marrow biopsy showing numerous amastigotes. Patient was treated with ketoconazole and rifamycin, but the patient died of treatment-related complications.[10] Soni *et al*. reported three cases of diffuse cutaneous and localized cutaneous leishmaniasis in HIV-infected patients which were unresponsive to parenteral and intralesional sodium stibogluconate.[11] Spontaneous healing of mucocutaneous leishmaniasis in HIV-infected patients is rare, but in immunocompetent patients, lesion heals in a period of 1 month to 3 years.[12] This was in contrast to our patient where significant reduction in the size of lesion was observed after 2 months of HAART and co-trimoxazole prophylaxis only. This may be due to increase in immunity or some role of co-trimoxazole against leishmania.

CONCLUSION

In endemic areas, leishmaniasis should be considered as a differential for nasal obstructive masses. Mucocutaneous leishmaniasis is uncommon in our region and any patient presenting with such disfiguring lesions should be investigated for any immunocompromised state. Furthermore, toxicity of anti-*Leishmania* drugs is more in HIV-infected patients; therefore, patients with mucocutaneous leishmaniasis on HAART and co-trimoxazole prophylaxis can be followed up for initial months for spontaneous improvement of the lesion.

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Conflicts of interest
There are no conflicts of interest.

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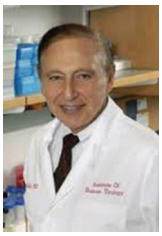

Robert Gallo

Robert Charles Gallo (born March 23, 1937) is an American biomedical researcher. He is best known for his role in the discovery of the human immunodeficiency virus (HIV) as the infectious agent responsible for acquired immune deficiency syndrome (AIDS) and in the development of the HIV blood test, and he has been a major contributor to subsequent HIV research.

Gallo was born in Waterbury, Connecticut to a working-class family of Italian immigrants.

In May 4, 1984, Gallo and his collaborators published a series of four papers in the scientific journal *Science*[14] demonstrating that a retrovirus they had isolated, called Human T-Cell Lymphotropic Virus-III in the belief that the virus was related to the leukemia viruses of Gallo's earlier work, was the cause of AIDS. A French team at the Pasteur Institute in Paris, France, led by Luc Montagnier, had published a paper in Science in 1983, describing a retrovirus they called LAV (lymphadenopathy associated virus), isolated from a patient at risk for AIDS.[16]

Gallo is the director and co-founder of the Institute of Human Virology (IHV) at the University of Maryland School of Medicine inBaltimore, Maryland Distinguished Professor in Medicine. Gallo is also a co-founder of biotechnology company ProfectusBioSciences, Inc. and co-founder and scientific director of the Global Virus Network (GVN).