Co-existence of Cutaneous Leishmaniasis with Pleural Effusion: A Case Report from Iran

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Abstract- Herein, a 12-year-old Afghan boy with chronic cutaneous leishmaniasis on the face and verrucous lesions on the body and pleural effusion suspected of having co-existent tuberculosis has been presented. The cutaneous lesions were appeared for five years before his admission. Leishman-Donovan bodies were seen in H&E (Hematoxylin and eosin) slide of skin lesion specimens. The pathogenic species was proved to be *Leishmania tropica* using Polymerase Chain Reaction (PCR) method. Purified Protein Derivative (PPD) and Leishmanin Skin Test (LST) were strongly positive. The patient was treated with systemic and intralesional meglumine antimoniate (Glucantime) for cutaneous leishmaniasis and then with anti-tuberculosis drugs for pleural effusion. Afterwards, pleural effusion was disappeared and cutaneous leishmaniasis cured.

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Keywords: Leishmaniasis; Cutaneous; Pleural Effusion; Tuberculosis; *Leishmania tropica*

Introduction

Chronic cutaneous leishmaniasis (CL) is mostly caused by *Leishmania tropica* infection in the Old World. Approximately 4% of *L. tropica* infections have been reported from Iran and Afghanistan. Keloidal and verrucous lesions are rarely reported on lower limbs (1).

In chronic leishmaniasis, the diagnosis is not easily made and amastigotes are rarely found in lesions. Polymerase Chain Reaction (PCR) method could be used for the diagnosis and species identification of chronic leishmaniasis (2).

Co-existence of American cutaneous leishmaniasis (ACL) with lepromatous leprosy and pulmonary tuberculosis has been reported (3). A case of disseminated cutaneous leishmaniasis with tuberculous dactylitis has also been reported from Iran (4).

In this case report, a 12-year-old Afghan boy with pleural effusion indicative of having tuberculosis and chronic leishmaniasis on the face, body, and extremities is reported.

Case Report

In September 2010, a 12-year-old Afghan boy with multiple erythematous nodular lesions on the face, verrucous and keloidal lesions on the right ankle and the right big toe, and edema of right foot referred to dermatology clinic of Razi Hospital, Tehran, Iran. Skin lesions were presented since seven years ago and were worsen in spring and summer.

At the time of admission, his medical history was negative for cough, sputum, chill, nocturnal sweating, and weigh loss. He had previously been treated with anti-tuberculosis drugs for facial lesions with the diagnosis of cutaneous tuberculosis in Afghanistan. Physical examination revealed dullness and decreased pulmonary sound in left costophrenic angle.

Multiple crusted nodules of the cheeks, forehead, chin, and ears were noted close to and inside the former CL scar (Figure 1). In addition, there were keloidal and verrucous lesions on the right lower leg accompanied with non-pitting edema (Figure 2).

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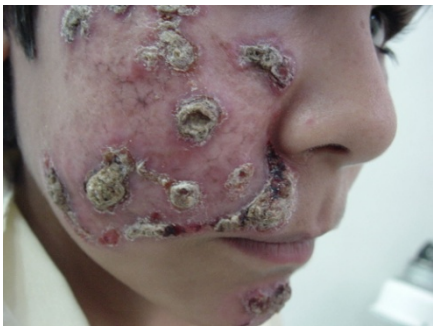

Figure 1. Crusted nodules on an erythematous base on right cheek

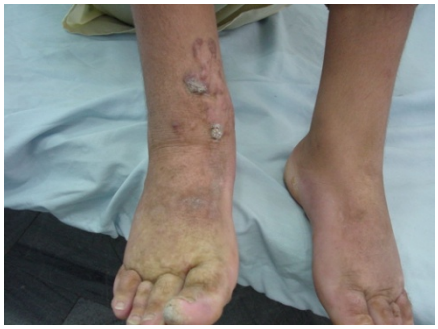

Figure 2. Keloidal and verrucous lesions on the right lower leg

Complete blood count, with differentiation, erythrocyte sedimentation rate, and other laboratory tests were within normal limit. Human Immunodeficiency Virus (HIV) test was negative. Pleural effusion of left lung was noted on chest x-ray (Figure 3).

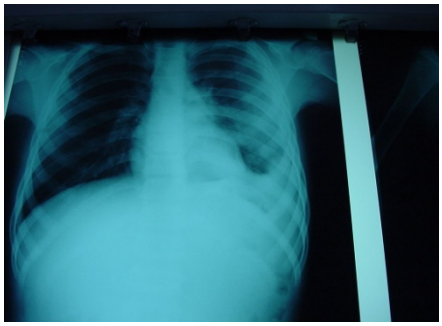

Figure 3. Pleural effusion of the left lung (Chest x-ray)

Lung CT scan showed increased pleural thickening accompanied by mild fibrotic changes probably in favor of tuberculosis (TB) (Figure 4). Purified Protein Derivative (PPD) and Leishmanin Skin Test (LST) were strongly positive. Smears and cultures of facial lesions for *Mycobacterium tuberculosis* and deep mycosis were negative. Direct smear of facial lesions showed Leishman-Donovan bodies (Figure 5). Polymerase chain reaction (PCR) was positive for leishmaniasis and *L. tropica* was confirmed as the causative agent. Indirect Immunofluorescence Antibody (IFA) test was positive with the titer of 1/640. Pleural effusion aspiration and lung biopsy were not performed due to patient refusal. Therefore, the diagnosis of TB was not confirmed.

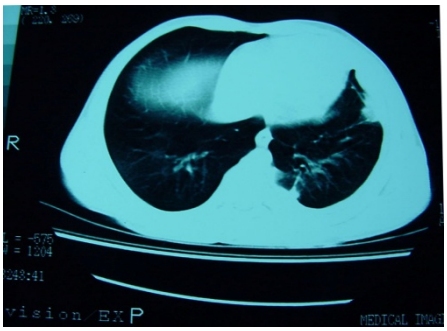

Figure 4. Lung CT scan showing increased pleural thickening

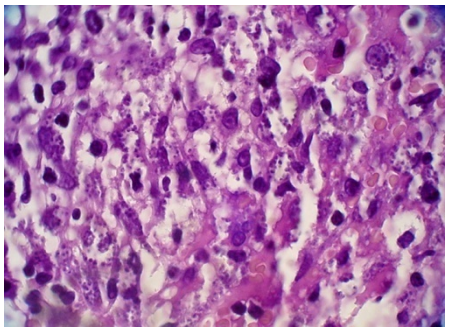

Figure 5. H&E slide of skin biopsy shows Leishman-Donovan bodies

Systemic (intramuscular) meglumine antimoniate (Glucantime), 20 Sb^{5+} mg/kg/daily, for 2 weeks and also intralesional were used for treatment of leishmaniasis. Then the patient was treated with standard four-drug anti-TB regimen (rifampicin, isoniazid, ethambutol and pyrazinamide) for pleural effusion for six months. Afterwards pleural effusion disappeared and cutaneous leishmaniasis lesions cured cosmetically acceptable.

Discussion

Co-existence of tuberculosis with visceral leishmaniasis, especially among HIV positive patients has been reported (5-7). However, the patient in this study was HIV negative. Positive LST and PPD showed that the patient is immunocompetent. Co-infection of visceral leishmaniasis, *Pneumocystis carinii* pneumonia, and pulmonary tuberculosis in a 29 year-old male HIV negative patient has been reported in Brazil (8). Moreover, in a 44-year-old male Brazilian immigrant, co-infection of American cutaneous leishmaniasis, lepromatous leprosy, and pulmonary tuberculosis has been reported. Immune assessment of this Brazilian

patient showed a Th1 type of immune response and good immune competency (3).

Visceral leishmaniasis-tuberculosis co-infection has been reported in East Africa. In a cross-sectional study conducted on active visceral leishmaniasis in Eastern Sudan, although LST and PPD were both positive among 20% of studied population (77 of 382 patients), there was no association between risk of infection with *L. donovani* and *M. tuberculosis* (7).

In another study, it has been mentioned that in spite of its low sensitivity, serology remains as a beneficial diagnostic tool, mainly for its high specificity and low cross-reactivity in patients co-infected with leishmaniasis and tuberculosis (9). In the present patient, the high titer of serologic tests could be explained due to chronicity of lesions and visceral involvement.

Although visceral leishmaniasis has become a frequent complication of HIV infection in many locations (10-11) co-infection of cutaneous leishmaniasis and pulmonary tuberculosis has been reported very rarely. A patient with disseminated cutaneous leishmaniasis with tuberculous dactylitis has been reported previously (4). In addition, a 52-year-old man with severe local destruction of his upper and lower lip and anterior nasal septum was diagnosed with mucosal leishmaniasis and tuberculosis lymphadenitis and inherent immune deficiency (12).

To the best of our knowledge, there are few reports of co-infections of cutaneous leishmaniasis and tuberculosis (3,12). Although we failed to confirm the diagnosis of pleural or pulmonary tuberculosis, radiographic study and response to anti-TB therapy were suggestive for tuberculosis. We decided to report this case because of its rarity.

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