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Disseminated Cutaneous Leishmaniasis in a Man With Human Immunodeficiency Virus Infection

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Introduction: Diffuse Cutaneous Leishmaniasis (DCL) is a rare form of Cutaneous Leishmaniasis (CL) observed in immunocompromised patients. This form has a chronic relentless course and is usually refractory to treatment.
Case Presentation: Here is the report of a 43-year-old man referred to Infectious Diseases Clinic in Boo-Ali Hospital (Zahedan City, Southeastern Iran) because of multiple chronic ulcers for five months. During the time, he had been visited by many physicians and received many different antibiotics, but there was no relief.
Conclusions: Although DCL is rare, physicians should mind its occurrence it in any immunocompromised patient with diffuse cutaneous ulcers refractory to treatment.

Keywords: Disseminated Cutaneous Leishmaniasis; HIV; Immunodeficiency

1. Introduction

Cutaneous Leishmaniasis (CL), an endemic disease in many countries around the world, is transmitted by the sand-fly and characterized by the development of cutaneous papules which progress into nodule then form ulcer and heal with permanent scarring (1-3). The disease is divided into the old and new world forms. The old world form is subdivided into urban and rural types and caused by organisms of *Leishmania tropica* complex. The new world form is caused by *L. Mexicana* and *L. viannia* complexes (1, 3). But, Diffuse Cutaneous Leishmaniasis (DCL) is a rare form of CL in immunocompromised patients such as patients with Human Immunodeficient Virus (HIV) or Acquired Immune Deficiency Syndrome (AIDS) whom the lesions are disseminated (at least ten ulcers) and it is characterized by a lot of clinical lesions including papules, nodules, and ulcers (2-6). Chronic form of Leishmaniasis caused by *Leishmania aethiopia* in Ethiopia and Kenya and various subspecies of *L. Mexicana* in the Central and South America are characterized by non-ulcerating, non-necrotizing skin lesions that spread over the body. This condition is associated with a suppressed cell-mediated immune response. Healing does not occur unless an acquired cellular hypersensitivity can develop (1, 4-7). Diffuse cutaneous forms have a chronic course and are usually refractory to treatment. Here in, is reported a 43-year-old man with AIDS who referred to the Infectious Diseases Clinic in Boo-Ali Hospital (Zahedan City, Southeastern Iran) because of multiple chronic ulcers for five months without any response to medical treatment.

2. Case Presentation

A 43-year-old man referred to the Infectious Diseases Clinic on September 2012 because of multiple chronic ulcers for five months without any response to medical treatment. He was from Saravan (A city in the Southeast of Iran in Sistan and Baluchestan province) where the incidence of HIV was higher than the other cities in this region (8). The first ulcer appeared five months ago on the wrist and it was a popular-nodular rash, which gradually spread and involved the entire forearm. Afterwards, several ulcers like the first one came into view on the left and right foot and then upper chest (23 ulcers). During this time, he had referred to many physicians and received a lot of different antibiotics, but there was no sign of healing. He was very thin without any other sign or symptom. He had worked for three years as a driver in Qatar 10 years ago and denied any sexual intercourse over there. According to laboratory report, his blood sample was positive for HIV infection with CD4 and CD8 counts of 75 cells/mm^3 and 699 cells/mm^3, respectively. Biopsy of the skin lesions revealed the Leishman bodies. Serology for Hepatitis B and C (HBV, HCV) infections, and VDRL test were negative; Electrocardiogram (ECG) was normal. Chest X-Ray showed a reticular infiltration in the right lobe, but he had no pulmonary complaint; purified protein derivative (PPD) test was negative; evaluation for active Tuberculosis was also negative. He was treated with sodium stibogluconate (SSG); 20 mg/kg/day for 30 days for Leishmaniasis, and then referred to HIV center to start antiretroviral therapy (ART) and follow-up carefully.

However, no side effects of SSG were observed during the treatment. The skin lesions decreased in size and the clinical response was good.

3. Discussion

Leishmaniasis is transmitted by the bite of infected female Phlebotomine sandflies. The sandflies inject infective promastigotes during blood meals. Promastigotes are phagocytized by macrophages and then transform into amastigotes form (1, 7-10). Amastigotes multiply in the infected cells and affect different tissues, depending in part on which Leishmania species are involved. Around 12 million people around the world are infected, with 1.5-2 million new cases every year. Disease can present in three forms: cutaneous, mucocutaneous, or visceral Leishmaniasis (1, 10). Cutaneous form occurs with skin ulcers, while the mucocutaneous form presents with the skin, mouth, and nose ulcers. The visceral form starts with skin ulcers and then accompanied with fever, enlarged spleen and liver and low red blood cells. Infection in humans is caused by more than 20 species of Leishmania. All the three forms can be diagnosed by observing the parasites under the microscope. Visceral disease can also be diagnosed by serology tests (1, 7, 9, 10). The treatment is determined by where the disease is acquired, the species of Leishmania, and the type of infection (1, 7, 10). Pentavalent antimony (sodium stibogluconate or meglumine antimonate) is used in CL. There are other drugs to treat CL including: liposomal amphotericin B; oral miltefosine; oral ketoconazole, itraconazole, and fluconazole are approved, but none is as effective as the pentavalent antimony compounds and topical paromomycin. Local therapies are also effective for some forms of CL including the following: cryotherapy and local heat therapy at 40 - 42°C. Other important issues in the management of Leishmaniasis are as follows: correction of malnutrition; treatment of concurrent systemic illness (HIV disease, Tuberculosis); control of local infections.

Disseminated CL is described in the Northern and Northeastern regions of Brazil. Treatment with Glucantime can cure patients with DCL. Many of the patients with disseminated CL have an immunodeficiency disorder (1, 7, 9). The patient under study had AIDS, but he presented multiple ulcers due to Leishmaniasis. Cutaneous Leishmais can be visible as an opportunistic infection associated with HIV/AIDS and may be the first symptom in patients with HIV infection in an endemic area (2-5, 8, 9). In 2013, a relationship between disseminated CL and CD4 count was reported by Mendes et al. (6). They showed that chronic inflammations in all DCL non-ulcerated lesions are predominantly formed by macrophages, plasmacytes, and T- and B-cells. Philips et al. (7) reported an immunocompetent patient diagnosed to have visceral Leishmaniasis along with simultaneous disseminated mucocutaneous and ocular involvement, a combination that was never reported before. Cases of Leishmania/HIV co-infections are reported in various parts of the world and it is no longer restricted to endemic areas. Several cases of HIV/Leishmaniasis co-infection are reported from Cameroon, Guinea Bissau, Mali, and Senegal. Leishmania/HIV co-infection is a serious problem (4, 5, 8, 9). A person with AIDS is more likely to develop Leishmaniasis after exposure to the protozoa; an opportunistic infection like visceral leishmaniasis or DCL, which can decrease the latency period between infection with HIV and the onset of AIDS (8, 9). AIDS increases the risk of visceral Leishmaniasis by 100 to 1000 times in endemic areas. Since both Leishmaniasis and HIV destroy the immune system, effective treatment for Leishmania is difficult under such circumstances (3, 4, 8, 9). Studies in Southwestern Europe show that pentavalent antimonial is initially quite effective with cure rates as high as 83%; however, 52% of the patients relapse between one and four times within a period of one month to three years. Alternative treatments are amphotericin B and lipid formulations of amphotericin B (3, 4, 8, 9). The current study patient could not be followed up more than seven months during which the ulcers were restricted and he received ART regimen. Although, DCL is a rare disease, physicians should mind its occurrence in any patient who is immunocompromised and also has multiple cutaneous ulcers - refractory to treatment.

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