Case Report

Coexistence of Recidivans, Mucocutaneous and Osseous Leishmaniasis in an Immunocompromised Patient

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Abstract

Leishmaniasis is a parasitic disease caused by the protozoan Leishmania, transmitted by female phlebotomine sandfly. The disease spectrum ranges from limited cutaneous plaques to disseminated nodules, and may involve viscera including liver, spleen and bone marrow. Concurrent occurrence of various patterns of the disease in a single patient is rare. Here, we report a patient with leishmania recidivans and mucocutaneous disease alongwith involvement of multiple bones, a combination that has not been reported in Pakistan.

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Introduction:

Leishmaniasis is caused by the genus Leishmania which is an obligate, intramacrophage, protozoan parasite. The vector responsible for the disease is sand-fly Phlebotomus. Along with the skin, the disease may involve mucous membranes and internal organs[1]. Commonly effected parts of the world include South Asia and Middle East. Cutaneous leishmaniasis is fur-ther divided into classical cutaneous leishmaniasis, diffuse cutaneous leishmaniasis, leishmania recidivans and post Kala-azar dermal leishmaniasis[2]. Visceral form of disease is of classical and viscerotropic type. Simul-taneous occurrence of cutaneous and visceral forms of disease in a single host is seldom reported and is a rare entity[3].

Case report

A 20 years old male from Hango, North of Pakistan, presented with recurrent multiple erythematous crus-ted plaques and nodules on face and limbs for last eight years. These plaques used to heal in a month to leave atrophic scars. Few plaques developed in old scars as well. He developed multiple plaques on forehead, left cheek and left nasal mucosa, which also healed with scarring and deformity. He also developed nodule in middle finger of right hand with pain and restricted movement of interphalangeal joints. He had pain in multiple bones and joints especially those with over-lying nodules, plaques and scars. He had weight loss and malaise. He had been on multiple medications includ-ing steroids, antimonials, antituberculous therapy and antibiotics, without relief. His sister too had similar lesions and was diagnosed as having ocular leishmani-asis.

On examination, he was well oriented, pale and was short for his age. He had hypertrichosis on face and body. He had reduced muscle mass on limbs. There were multiple crusted nodules and plaques on shoulder-s, knees and left foot (Figures 1-4). He had a firm subcutaneous nodule involving the middle finger of his right hand (Figure 3). It was immobile, tender and adhe-rent to underlying structures. The movements of the finger were restricted. He had onychomycosis of multi-ple nails of his fingers and toes and pityriasis versicolor on trunk and limbs. There were multiple atrophic depressed scars on face, shoulders, arms, hands, knees,

legs and feet (Figure 5). Cervical lymph nodes were palpable. Many of his bones and joints were tender including shoulders, left knee and left shin. Rest of systemic examination was unremarkable and no visceromegaly was noted.

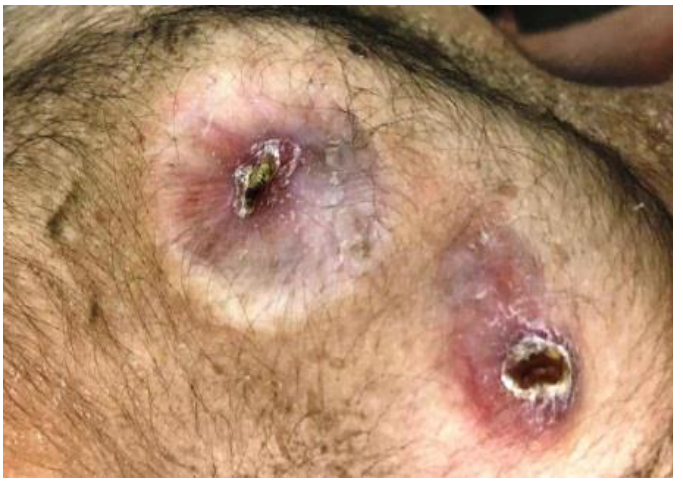

Figure 1: *Plaques on old scars on left shoulder.*

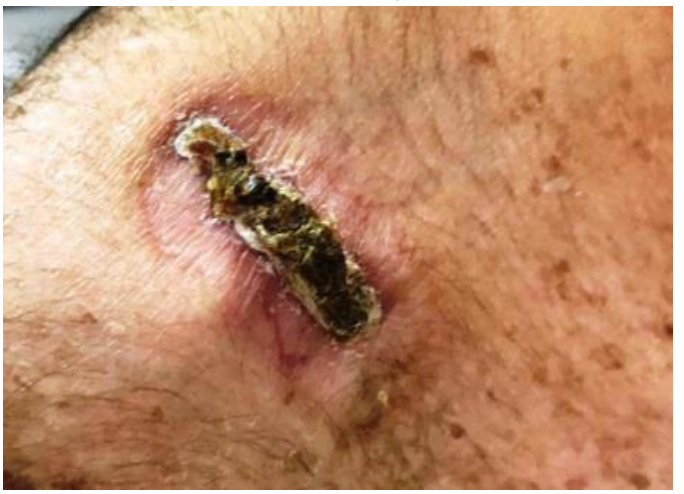

Figure 2: *Crusted plaque on right shoulder*

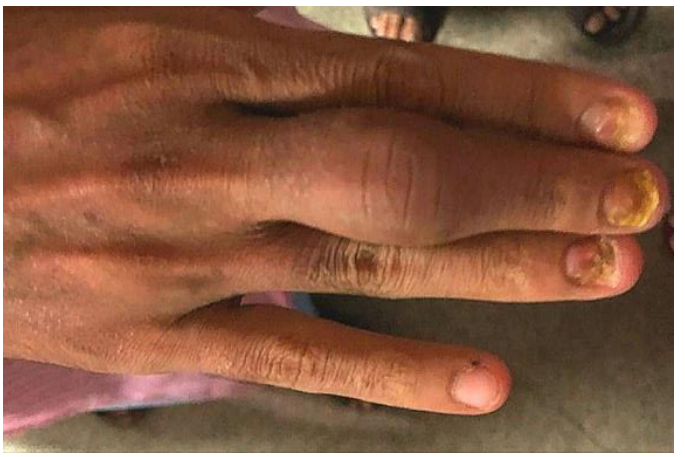

Figure 3: *Right hand showing onychomycosis of nails and subcutaneous nodule involving middle finger.*

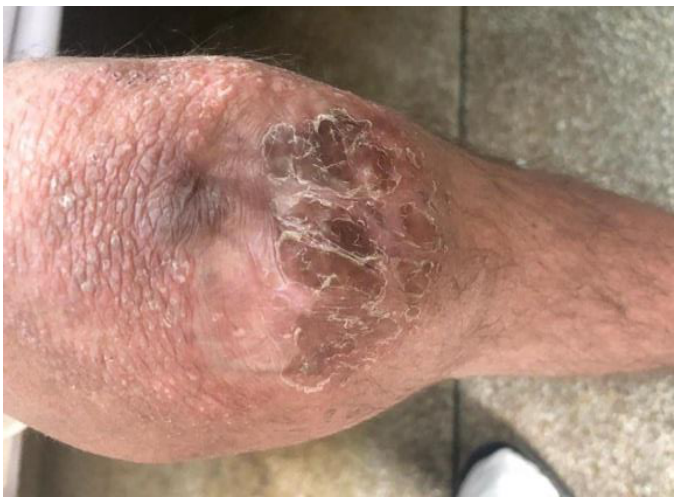

Figure 4: *Crusted plaques on left knee*

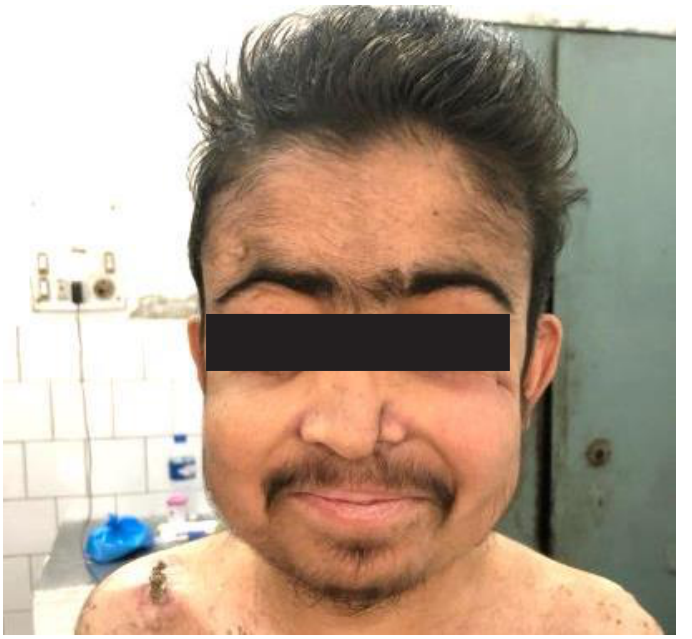

Figure 5: *Old scars of healed lesions on face, involving underlying nasal mucosa)*

His investigations showed hemoglobin of 9.4 g/dL, a total leucocyte count of 20,400/mm^3, platelet count 713,000/mm^3, an ESR of 70 with microcytic, hypochromic anemia, alkaline phosphatase was 764 U/L, LDH was 876 U/L , serum IgG level was 1775mg /dL, anti-HIV, HBsAg and anti-HCV were negative. His sputum and smear from cutaneous lesions were also negative for tuberculous bacilli.

Smears taken from cutaneous nodules on shoulders demonstrated significant number of LD bodies when stained with Giemsa (Figure 6).

On histopathological examination of nodule in finger, there were numerous necrotizing granulomas. Most of

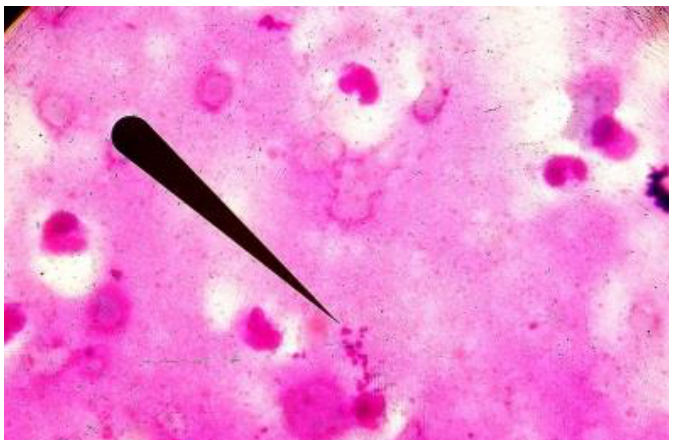

Figure 6: *Smear from crusted nodule showing LD bodies stained with Giemsa.*

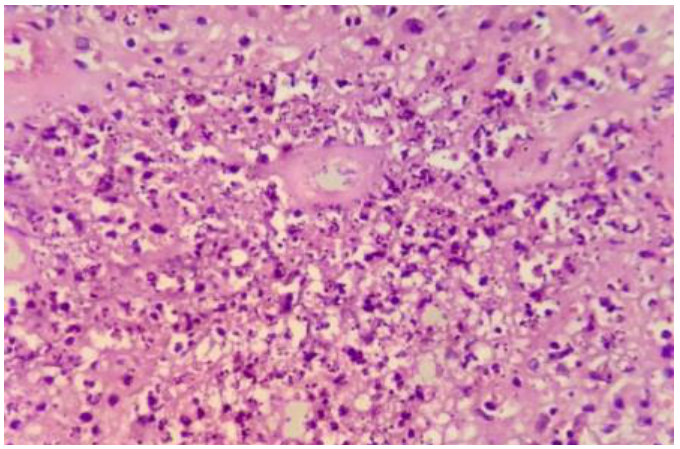

Figure 7: *Histopathology (H & E stain) of nodule showing necrotizing granulomas and LD bodies (free and within histiocytes)*

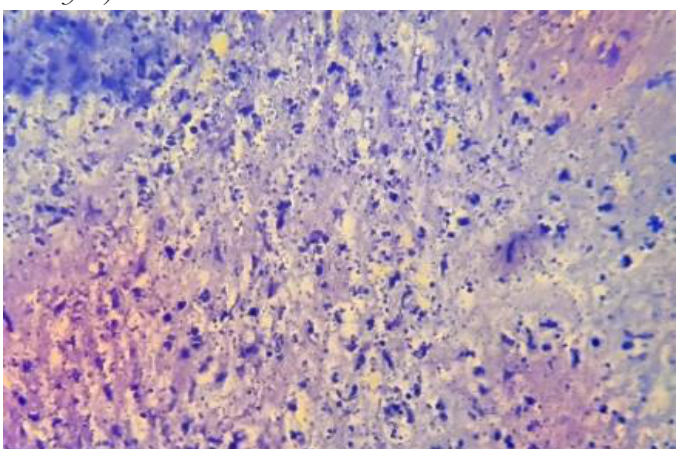

Figure 8: *LD bodies highlighted by Giemsa stain*

them had lots of viable as well as degenerating neutrophils at the center. In addition, there were numerous LD bodies scattered all over. These were present within histiocytes as well as free within the tissue (Figure 7). Several degenerating bony spicules were also seen hinting at bone damage. Giemsa stain highlighted the LD bodies even further, which are seen as round to ovoid bodies measuring 3x1 to 6x3 μm (Figure 8).

Radiological investigations revealed soft tissue swellings and lytic lesions in bones along with obliteration of joint spaces, consistent with cutaneous and skeletal manifestations of leishmaniasis (Figures 9-11). However, chest radiograph and abdominal ultrasound were normal.

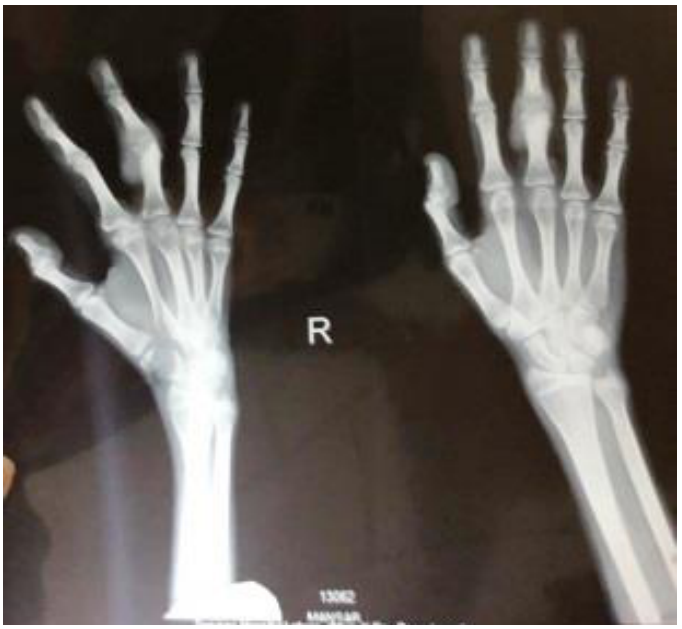

Figure 9: *Multiple lytic and degenerative lesions in the proximal and middle phalanges of the middle finger and carpal bones*

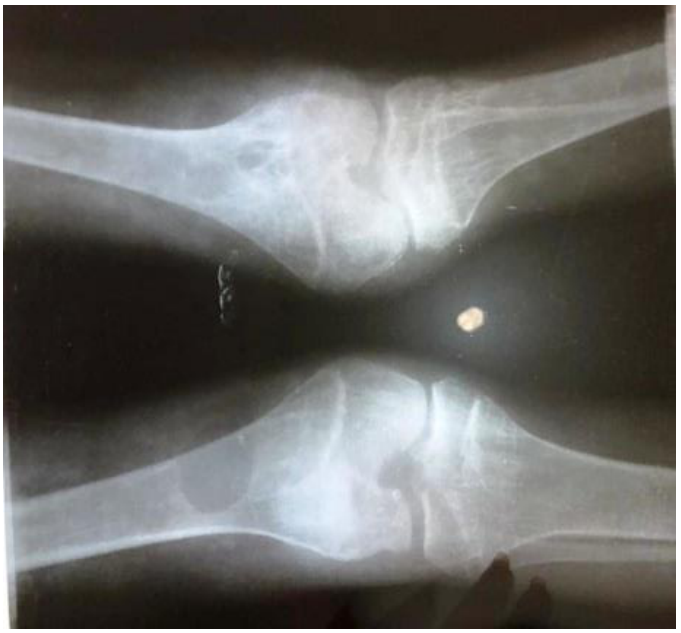

Figure 10: *Multiple with marked osteopenia in bilateral distal femora*

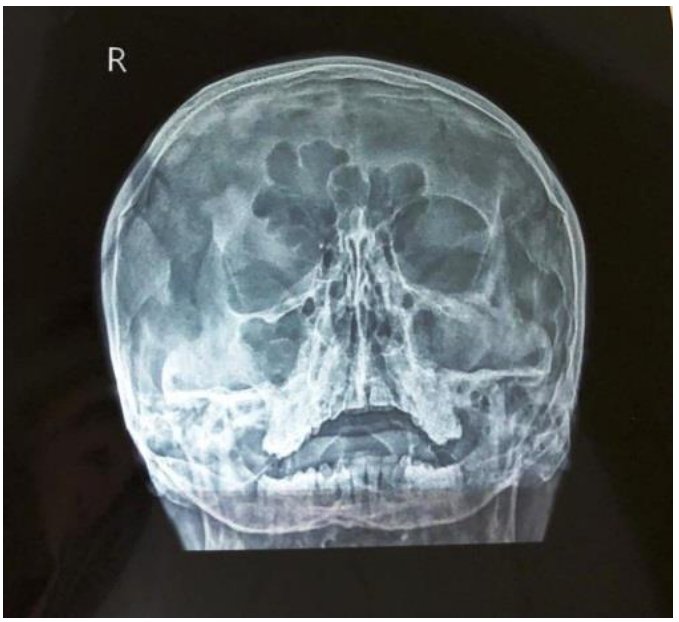

Figure 11: *Lytic lesions in skull*

Figure 9: Multiple lytic and degenerative lesions in the proximal and middle phalanges of the middle finger and carpal bones. Absence of periosteal reaction, marked osteopenia and reduced interdigital spaces along with soft tissue swelling around proximal interphalangeal joint of middle finger and increased soft tissue haze in lateral half of right hand. Multiple lytic lesions with marked osteopenia in bilateral distal femora (Figure 10) and skull (Figure 11)

Discussion:

Based on the clinical features, other diagnostic considerations must include cutaneous tuberculosis, atypical mycobacterial infection, deep fungal infection, sarcoidosis and multiple myeloma. These were ruled out on histopathology, cultures and serology (Bence Jones proteins were also negative). Being an endemic area, cutaneous and visceral leishmaniasis has been reported from Pakistan.[4] However, osseous involvement in the form of lytic lesions involving whole body has not been reported. Two patients have been reported to have osseous Leishmaniasis involving bones of distal extremities from Brazil[5]. Another patient with involvement of femur has been reported from Iran[6]. Our patient had involvement of multiple bones including skull and bones of limbs. Another interesting feature was presence of ocular leishmaniasis in the sibling. Leishmaniasis can have a myriad of histopathological features. The most common and diagnostic of these is granulomatous inflammation. These granulomas themselves can be of various types viz sarcoidal granulomas, necrotizing granulomas, suppurative granulomas and palisaded granulomas[7]. A mixture of patterns may be seen in a single case[8]. Our case showed necrotizing as well as suppurative granulomas along with bony spicules. Leishmaniasis can affect and damage a variety of organs in the body[9]. The patient was immunocompromised as he had onychomycosis and pityriasis versicolor. Atypical forms of leishmaniasis have been reported in immunocompromised patient[10]. We demonstrated LD bodies along with bony spicules indicating bone involvement with leishmaniasis, a rarely reported entity. Another striking feature noted was presence of multiple patterns of disease in a single patient. This highlights the possibility of changing pattern of disease, microbial characteristics and ultimate manifestations in host.

Limitation

Facilities for Leishmanial culture are not available in Pakistan, therefore we couldn't know more about the species of the parasite involved.

Conclusion:

Extensive work up in this case helped us to reach the diagnosis of cutaneous leishmaniasis with bone involvement. So, evaluation of bone lesions by radiographic examination seems to play an important role in identification and management of these lesions.

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