CASE REPORT

Localized Cutaneous Leishmaniasis presenting in a sporotrichoid pattern: A Case Report

Jasem Yousef Al Sanea, MD

Department of Dermatology, Amiri Hospital, Kuwait

ABSTRACT

Leishmaniasis is a major infection worldwide, affecting more than 95 countries in five continents. It is caused by Leishmania parasites through the bite of infected female sand fly. The clinical manifestation of leishmaniasis ranges from cutaneous ulcers to systemic multiorgan disease. Localized Cutaneous Leishmaniasis (LCL) is characterized by pink-colored papule that enlarges to nodule or plaque-like lesion, with painless ulceration, white-yellow crust, and indurated border. It is the most common form of cutaneous leishmaniasis, as it has many clinical forms. Here, I report a case of a 45 years old Indian male, living in Failaka island, who presented with linearly arranged localized cutaneous leishmaniasis lesions on right upper extremity, of one month duration. Early diagnosis and treatment is extremely important to prevent complications and further spread of the infection.

INTRODUCTION

Leishmaniasis is transmitted by the bite of infected female phlebotomine sandflies to a mammalian reservoir, typically rodents, sloths, and marsupials. Sandfly injects promastigotes into the skin through a blood meal, which is then phagocytized by macrophages and transformed to amastigotes. Sandfly takes a blood meal of the macrophage-infected amastigotes and transform them back to promastigote in their midgut, where they divide and migrate to proboscis. Transmission occurs when a sand fly habitat is disturbed, or when susceptible hosts move into areas of endemic transmission. However, the global prevalence of cutaneous leishmaniasis is increasing due to mass population displacement, war, climate change, and adventure travel. Leishmania causes a spectrum of cutaneous disease, ranging from mucousal leishmaniasis and leishmaniasis recidivans, which are caused by an oligoparasitic disease, to the poly parasitic diffuse cutaneous leishmaniasis. The most common form is the localized cutaneous leishmaniasis, which tends to occur on exposed areas of the skin with an incubation period of few weeks to months. Localized cutaneous leishmaniasis clinically begins as a pink-colored papule, that enlarges and develops into a nodule, with a painless ulcerative center. The ulcerative center can be covered with hyperkeratotic eschar or with thick white-yellow fibrous material depending on species. It can present in different clinical pattern which includes sporotrichoid, verrucous, zosteriform, psoriasiform, or eczematous features. Spread of lesions along the draining lymphatics may also be visualized and palpated subcutaneously. Localized Cutaneous leishmaniasis gradually heals over months to years, depending on the species.
and size of the lesion. Healing may be with an atrophic, depressed, or keloid scarring. L. tropica infection is usually more chronic species, with relatively small (1-2 cm) lesions that may be larger on the face.

**CASE REPORT**

A 45 years old Indian male presented to the outpatient clinic with multiple well-defined, painless, non-pruritic nodule on the dorsal aspect of right hand, elbow, and extensor aspect of right forearm of one month duration. These nodules had an ulcerative center associated with crusting and followed the pattern of lymphatic drainage. [Fig. 1,2,3] Cutaneous examination showed well-defined plaque-like lesion with central ulceration, around 3 cm X 3 cm in size with regular borders. [Fig. 1, 2] They started as pink-colored papules that increased progressively in number and size. There was no sign of regional lymphadenopathy. He did not complain of nasal symptoms, denied recent travel history, or any history of immunosuppressive therapy. The patient lived in Failaka island, which is known to be endemic for L. tropica. The patient had uncontrolled hypertension and diabetes mellitus, for both of which he was not taking any medications. We kept a differential diagnosis of sporotrichosis, atypical mycobacteria, pyoderma gangrenosum, and ecthyma. Punch biopsy was taken and it showed skin with dermis showing acute-chronic inflammatory cell infiltrate. [Fig. 4]. Few histiocytes showed amastigote form of leishmania in the cytoplasm. [Fig. 5] Giemsa staining showed blue cytoplasm, violet-blue nucleus, and the kinetoplast red to violet (a critical diagnostic characteristic). [Fig. 6].
Localized Cutaneous Leishmaniasis presenting in a sporotrichoid pattern: A case report

DISCUSSION

According to World Health Organization (WHO) report, the annual incidence of leishmaniasis is estimated to be 0.7 to 1.2 million new cases per year. Cutaneous leishmaniasis poses a major health problem in countries like Afghanistan, Pakistan, Algeria, Syrian Arab Republic, and Brazil. Leishmaniasis can have various cutaneous to visceral manifestations. The mode of spread is through the bite of infected female phlebotamine sandflies on an exposed skin. Leishmania infect the macrophages of mammalian host tissues in their amastigote form. They develop into motile, flagellated promastigote in the gut of the sandfly. Promastigote parasites are then regurgitated from the sand fly into the next mammalian host. Preventive measures to reduce infection and transmission include covering the skin with clothing, application of skin repellents, and usage of permethrin as an insecticide. The wide clinical spectrum of cutaneous leishmaniasis may be due to variability in parasite virulence and variability in host immune response. Localized cutaneous leishmaniasis is the most common form of cutaneous leishmaniasis, which tends to occur on exposed areas of the skin. At least 23 species have been associated with human infection, which can be subcategorized into New-World Leishmaniasis and Old-World Leishmaniasis. Presentation varies from pink-colored papules to nodules, single to multiple lesions, hyperkeratotic eschar ulcer, to thick white-yellow fibrinous ulcer. The disease may also follow sporotrichoid, verrucous, zosteriform, psoriasiform, and eczematous features. Our case presented as multiple nodules with an ulcerated centre associated with crusting with a distribution that follows sporotrichoid pattern. In fact, the pattern of distribution suggested that we could add Tuleremia as a differential diagnosis. However, the main reason we considered leishmaniasis as a provisional diagnosis was general painlessness, occurrence on an exposed location, and presence of inflammatory satellite papules, as well as subcutaneous induration beneath lesion, and ulcerations with well-defined indurated borders. Diagnosis of cutaneous leishmaniasis is at times difficult of because the clinical appearance of the lesion can mimic other diseases. Thus, investigations (histopathology, culture, molecular techniques, tissue gram stain) are needed. At times, culture may not yield positive results because organisms may be scarce especially in leishmaniasis recidivans and L.V. braziliensis. In our case, histopathology with its characteristic findings were enough to confirm the diagnosis.

Fig. 5 Showing histiocytes containing amastigote form of leishmania.

Fig. 6 Giemsa stain showing histiocytes containing amastigote form of leishmania with blue cytoplasm, violet blue nucleus and red to violet kinetoplast.
Visualization of the rod-shaped kinetoplast and characteristic appearance of LD bodies on giemsa stain, with the red to violet kinetoplast, as well as blue cytoplasm and violet blue nucleus confirmed the provisional diagnosis. We took a punch biopsy on the edge of the lesion to eliminate the element of pus and necrotic debris that may interfere with the characteristic findings and it revealed dermis showing acute-chronic inflammatory cell infiltrate, and few histiocytes show amastigote form of leishmania in the cytoplasm. Initial treatment includes local therapy (pentavalent antimonials and paromycin), followed by oral systemic therapy (azoles and miltefosine) and parenteral systemic therapy (pentavalent antimonials, amphotericin, and pentamidine) mostly for up to three weeks depending on the organism and the clinical manifestation.10,11

CONCLUSION
In conclusion, the diagnosis of localized cutaneous leishmaniasis is based on clinical features, skin biopsy for histopathology, culture, and tissue giemsa stain, as well as molecular techniques, and serology in some institutions. However, serology is less diagnostic due to the high yield of result in tissue histopathology and giemsa stain. In my case, as it is a rare presentation in Kuwait, we have to always keep in our minds the uncommon diseases.

ACKNOWLEDGMENT
Pathology Department, Amiri Hospital
Dr. Mohammed Al Enzi
Dr. Tamer Amer

REFERENCES