Case Report

Cutaneous Leishmaniasis in a Traveler Returning From Sri Lanka

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Abstract

Infection with protozoan parasites of the genus *Leishmania* causes a variety of clinical diseases called leishmaniasis, which is transmitted to human host by the bite of a sandfly. Here, we present a Taiwanese man with cutaneous leishmaniasis after he had traveled to Sri Lanka. A 35-year-old man had a painless erythematous nodule with central ulceration on the ventral side of the right wrist. The skin lesion developed 3 weeks after he returned to Taiwan from Sri Lanka to rescue tsunami sufferers. Excisional biopsy revealed dense and diffuse infiltration of histiocytes and lymphocytes throughout the dermis, and numerous amastigotes were identified in the cytoplasm of the histiocytes. Nine months after he was first treated, his skin lesion recurred at the same location. Under the diagnosis of cutaneous leishmaniasis, he underwent a second excision and took 400 mg itraconazole per day for 4 months. No further recurrence or internal organ involvement was noted. *(Tzu Chi Med J 2008;20(2):147–149)*

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1. Introduction

Leishmaniasis is caused by the protozoan *Leishmania*. The protozoan is transmitted to mammals, including humans, by the saliva of sandfly. It causes several diverse clinical diseases, including disseminated visceral infection (kala azar), cutaneous skin lesion, and mucosa leishmaniasis. Cutaneous leishmaniasis (CL) is the most common clinical type [1]. Taiwan is not an endemic area for leishmaniasis, but many Taiwanese travel throughout the world. Physicians should be aware of the possibility of contracting CL in travelers who have been to endemic areas. We herein present CL in a Taiwanese man who had returned from Sri Lanka.

2. Case report

A 35-year-old man volunteered to rescue sufferers in Sri Lanka after the tsunami in South Asia in December 2004. An erythematous papule appeared on the ventral side of his right wrist 3 weeks after he returned to Taiwan. Neither pain nor itching was noted. Gradually, it became an erythematous nodule with a central ulceration (Fig. 1). The nodule was treated with liquid nitrogen with poor response at a clinic. He was then referred to the Department of Dermatology at the Buddhist Tzu Chi General Hospital. Physical examination did not reveal any systemic symptoms or signs and the skin lesion was totally excised. Pathologic
examination revealed dense and diffuse infiltrate of histiocytes and lymphocyte in the dermis (Fig. 2A). On high magnification, the cytoplasm of the histiocytes were found to be filled with numerous bluish to grayish colored oval amastigotes (Fig. 2B). The diagnosis was CL. Because of complete skin recovery, he was lost to follow-up. However, 9 months after the initial surgery, an erythematous nodule recurred at the same location. It progressively enlarged with a central ulceration that produced a volcano-shaped lesion. He visited us and received a second excisional biopsy, which revealed a similar lobular granulomatous inflammatory pattern as that of the first biopsy, but with fewer amastigotes in the histiocytic cytoplasm. He received 400 mg itraconazole per day for 4 months after the second excision. No local recurrence or internal organ involvement was noted thereafter.

3. Discussion

The World Health Organization has estimated the prevalence of CL to be 12 million cases per year. CL is classified into two different clinical syndromes: New World, when it is acquired in the Americas, which is infected by \( L. \) \textit{mexicana} complex (\( L. \) \textit{mexicana}, \( L. \) \textit{mexicana} \textit{amazonensis}, \( L. \) \textit{mexicana} \textit{venezuelensis}); and Old World, when it is acquired in Asia, Africa, the Middle East, or Europe, which is infected by \( L. \) \textit{major}, \( L. \) \textit{tropica}, or \( L. \) \textit{aethiopica} [2,3]. Both the Old World and New World forms of CL present as a spectrum of diseases ranging from single, chronic ulcerative lesions (oriental sores) as in our case, to disseminated nodular lesions (diffuse CL). The typical lesion appears as an erythematous papule at the site of inoculation which increases in size and ulcerates. The lesions are round with raised borders and appear 2–8 weeks after the patient has been bitten by an infected sandfly. Outbreaks of CL may occur in war-torn countries or in areas after disasters such as the tsunami in the affected regions of Sri Lanka. It is not considered to be endemic in Taiwan and physicians in industrialized countries tend to have little experience in this type of diagnosis.

The first autochthonous case of CL in Sri Lanka was reported in 1992 [4]. The case incidence has increased and more than 600 cases of CL were reported in the past 4 years [5]. The pathogen of CL in Sri Lanka is \textit{Teishmania donovani} which is usually associated with visceral leishmaniasis in all Asian and Eastern countries [6,7]. This may require epidemiologic study to identify genetically susceptible patients.

Leishmanial infections are transmitted via the bite of infected female sandflies. The parasite lives as an extracellular, flagellated promastigote in the gut of the insect. After multiplication and differentiation in the

**Fig. 1** — An erythematous nodule with elevated borders and central ulceration on the ventral side of the wrist.

**Fig. 2** — (A) There are lobular granulomas with infiltration of lymphocytes, macrophages and epithelioid cells throughout the dermis; the margin is free (hematoxylin & eosin, 12.5×). (B) Numerous bluish, round to oval shaped amastigotes can be identified in the parasitized macrophages (arrows) (hematoxylin & eosin, 1000×).
sandfly gut, in approximately 1 week, the infectious promastigotes migrate to the proboscis. Following inoculation into the skin of a mammalian host, macrophages engulf promastigotes into the lysosomes and the parasites are transformed to obligate intracellular life, amastigotes. Free amastigotes are released from the infected macrophages and infect dendritic cells. Interleukin-12 is released from infected dendritic cells and the infected hosts generate antigen-specific T cell-dependent immunity, the interferon-γ-producing Th1 cells (8). An important characteristic of *Leishmania* infection is the presence of persistent parasites following resolution of the infection. It appears that these parasites are maintained long-term at a very low level by the immune response. As infection with *Leishmania* induces a strong cell-mediated immune response, it is unclear why the parasites are not completely eliminated. Recent studies indicated that one contributing factor may be the generation of regulatory T cells during the infection, which limit the immune response sufficiently to maintain persistent parasites. These regulatory T cells function in part by the production of interleukin-10 (9,10).

The clinical and histologic appearance of CL skin lesions is related to an interaction between the host’s immune response and the virulence factors of the different *Leishmania* species. The indication for systemic treatment is the presence of mucosal lesions, lymph node metastasis or lesions unresponsive to local treatment (11). The therapeutic cornerstone is *pentavalent antimonials* (Pentostam, Glaxo-Wellcome, Research Triangle Park, NC, USA) and meglumine antimoniate (Glucantime, Rhone-Poulenc, Paris, France), which remain the mainstay of therapy for most forms of leishmaniasis. However, those drugs are not available in Taiwan. Other major treatments include physical modalities (heat, cryosurgery), local or intraslesional injections, various anti-infective agents (dapsone, metronidazole, trimethoprim-sulfamethoxazole), amphotericin B, pentamidine, allopurinol, azoles, immunotherapy (i.e. interferon-γ), and a variety of agents (12). In our patient, CL presented as a local recurrent ulcerative nodule. He had tried cryotherapy first with no effect, then received surgical excision. Although recurrent, the amount of amastigote in pathology was reduced. After the second excision, he took itraconazole as combination therapy. Until this writing, no local recurrence or remote infection has been noted.

**References**