



Case report

Case report and literature review: Genital leishmaniasis

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ABSTRACT

The majority of genital ulcers are caused by sexually transmitted infections, though there are also other infectious and noninfectious etiologies. We present here an unusual cause of a penile ulcer due to cutaneous leishmaniasis, along with a review of the literature regarding such cases. The patient recovered following timely initiation of treatment. Rapid diagnosis of this case was aided by occurring in the context of a concurrent outbreak of cutaneous leishmaniasis in the region in which the patient resided

Introduction

The most common etiologies of genital ulceration are infectious, generally sexually-transmitted infections (STIs). There are also non-infectious causes ranging from trauma to autoimmune disorders to neoplasms. We present here an unusual infectious cause of genital ulceration.

Case presentation

A 36-year-old man, a construction worker, presented with a persistent, nonpainful ulcer on the shaft of his penis. Approximately 40 days before seeking care, a macular lesion appear on dorsum of his penis, accompanied by a similar lesion on his left buttock (Fig. 1). The lesions gradually enlarged, and the patient was able to express purulent discharge from them. The patient had no prior episodes of these symptoms, no fever, no weight loss or other systemic symptoms. He had no prior history of STIs; he was married and had no new sexual partners. His wife did not have any similar symptoms. Prior to seeking care he had not tried any form of self-treatment. He recently worked on a construction project, during which he slept outdoors at the site wearing only loose shorts and without any mosquito net.

On examination, a 1.2 cm ulceration was seen on dorsolateral aspect of penis. There was also another 2.3 cm lesion with satellite pustules on left buttock, but it was not in a sacral dermatome distribution. There was no inguinal adenopathy.

Complete blood count, CRP, and liver function tests (LFT) were normal. VDRL, HIV, and HSV serologies and Tzanck tests were negative.

Patient declined biopsy. However, Giemsa staining of the expressed purulence showed Donovan bodies in histocytes (Fig. 2).

The clinical appearance, along with the Giemsa findings, coupled with his overnight exposure to arthropod bites in the context of local epidemiological data was suggestive of leishmaniasis. Empiric treatment was started, monitoring LFT and electrocardiography for potential toxicity.

Treatment consisted of 20 mg/kg/day intramuscular meglumine antimoniate for 20 days. Intralesional medication was not used at the patient's request, as he was fearful of possible side effects on the penis. The patient discontinued treatment for three days due to mild medication intolerance, then resumed therapy with his physician's encouragement. After completing the full course, the lesions showed marked improvement, ultimately resolving completely. To our knowledge, no other co-workers on the site developed Leishmaniasis, though no extensive case-finding efforts was undertaken.

Discussion

The majority of penile ulcers are caused by STIs, including HSV, syphilis, lymphogranuloma venereum, granuloma inguinale, and chancroid. There are other bacterial and fungal infectious causes, including cutaneous tuberculosis. Behcet syndrome, Wegner granulomatosis, psoriasis, sexual trauma, and neoplasia are among non-infectious etiologies [1]. Syphilitic chancres are persistent and painless while HSV cause painful lesions. Some bacteria like *Haemophilus ducreyi* causes "soft chancre" with tender inguinal lymphadenopathy and painful ulcers. In this case, lesions were progressive and painless and adenopathy

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Fig. 1. Penile lesion (top) and buttock lesion (bottom) at time of diagnosis.

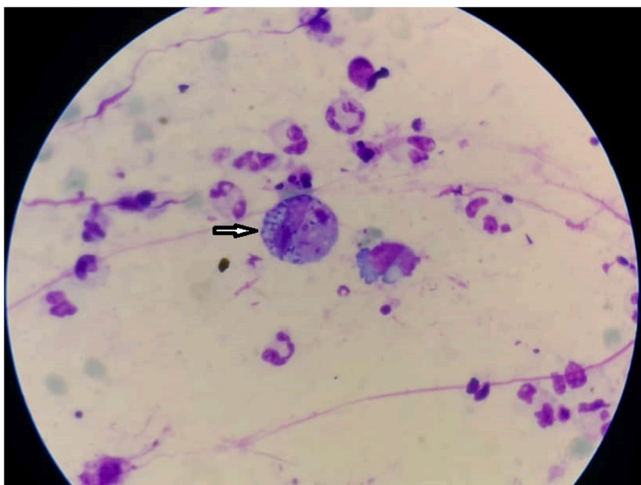


Fig. 2. Amastigotes in a histocyte (arrow).

was absent.

Leishmaniasis is a parasitic infection caused by a flagellated parasite belonging to the genus *Leishmania*. In most cases, it is a zoonotic disease transmitted via a bite by nocturnal hematophagous sand-flies of the genus *Phlebotomus*. The disease reservoirs consist of wild or semi-domesticated animals, generally rodents or dogs. The disease itself is distributed extensively worldwide in the Americas, Asia, Europe and Africa. Three major clinical forms are seen: cutaneous leishmaniasis, mucocutaneous leishmaniasis and visceral leishmaniasis [2]. Ninety

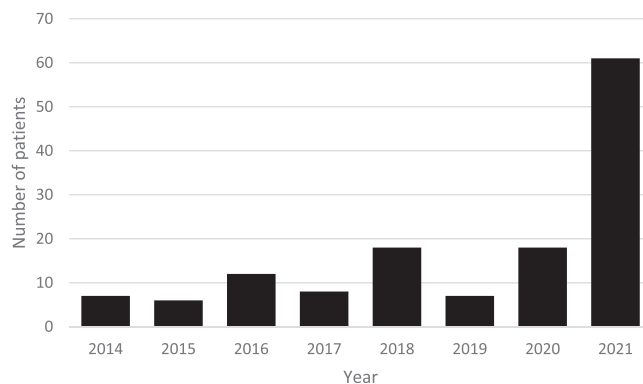


Fig. 3. Number of case of cutaneous leishmaniasis diagnosed in Sevah, Iran, and surrounding suburbs (population ~ 260,000).

percent of leishmaniasis cases are cutaneous. Cutaneous leishmaniasis is more frequently seen on exposed body areas such as the face, eyelids, forehead, hands, wrists and, occasionally, the legs. The involvement of the genitals is rare, although there are previous reports of infection of the penis [3–9]. The ages of those so infected range from infancy to octogenarians [10,11]. Generally, though not always, these lesions in areas normally covered by clothing are associated with immunosuppression or attributed to hematogenous spread in the context of multiple cutaneous lesions. Neither of these situations was true of the present case. In this patient, the loose-fitting night time clothing likely allowed vector access to the affected sites.

The diagnosis of cutaneous leishmaniasis is considered based on clinical appearance and a history of residence or travel in endemic regions. Diagnosis is confirmed by pathological, serological, or molecular-based testing. Leishmaniasis is endemic in the region in which this patient lived and worked, becoming more common in recent years (Fig. 3). Because of this, cutaneous leishmaniasis was considered early on, despite its unusual location. In addition to cutaneous leishmaniasis, post kala-azar dermal leishmaniasis (sequela of visceral leishmaniasis) has parasite-containing lesions that may be found a variety of skin and mucosal locations, including the genitals [12,13]. This condition was not present in the patient.

The presence of infection disease on the genitals raises the question of the potential for sexual transmission. There have been no reports of sexual transmission of cutaneous leishmaniasis in humans. However, there has been a single reported case of probable sexual transmission of the visceral form of the disease in humans [14]. Reports in the veterinary literature supports this possibility: reports of visceral leishmaniasis in canines demonstrate parasites in genital tissues and semen of male dogs capable of sexual transmission of leishmaniasis to females in the absence of the vector [15–18]. In a murine model, female-to-male transmission of visceral leishmaniasis has also been demonstrated [19].

This is, to our knowledge, only the second reported case of genital cutaneous leishmaniasis in Iran [20].

CRediT authorship contribution statement

Sasan Gazerani: Data curation, investigation, Project administration, Writing – original draft. **Mark K Huntington:** Conceptualization, Data curation, Investigation, Project administration, Supervision, Writing – review & editing. **Javad Satvati:** Data curation, Project administration.

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Ethical approval

This case report got ethical approval in Saveh University of Medical Sciences on 2022.06.19. approval ID is IR.SAVEHUMS.REC.1401.001.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflicts of Interest

I, Dr. S Gazerani, as corresponding author of this manuscript declare that there is no conflict of interests in this article.

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