An Unusual Case of Ectopic Tungiasis With Pseudoepitheliomatous Hyperplasia

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Tungiasis is caused by the penetration of the female sand flea *Tunga penetrans* into the epidermis, and subsequent hypertrophy of the parasite. In most cases lesions are confined to the feet. During a cross-sectional study, an unusual case of ectopic tungiasis in the inguinal area was detected. Histological examination of tissue samples showed a remarkable pseudoepitheliomatous aspect of the epidermis. Clinical features and differential diagnoses are discussed.
Key Words: Ectoparasite, *Tunga penetrans*, tungiasis, ectopic localization, histopathology, Brazil.

Tungiasis is a parasitic skin disease caused by the penetration of the female sand flea *Tunga penetrans* into the epidermis, with subsequent hypertrophy of the parasite. Beside man, various domestic and peri-domestic animals are affected [1]. The sand flea originated on the South American continent and the Caribbean Islands, but it was accidentally introduced into sub-Saharan Africa in the late 19th century [2]. It is endemic in many countries in Latin America, the Caribbean and sub-saharan Africa [3]. In Brazil, prevalence rates reach up to 60% in children in the miserable squatter camps at the outskirts of the big cities as well as in the under-developed rural hinterland [4-6].

In endemic areas, particularly in children, tungiasis is associated with severe morbidity [7]. Without appropriate treatment, secondary infections with pathogenic bacteria are extremely common [8]. In severe cases, lymph edema, deep ulceration with denudation of bones, gangrene and auto-amputation of digits can occur [3,9,10]. Tungiasis also has been associated with tetanus in non-vaccinated individuals [11].

As the sand flea is unable to jump high, it is generally assumed that lesions are confined to the feet. This, in turn, causes physicians to restrict their attention to the lower extremities. In order to determine whether tungiasis may occur at other sites than the feet, a cross-sectional study was carried out in a population living in a highly endemic area in a slum in Fortaleza, Northeast Brazil. During the systematic screening, a very particular case of tungiasis was detected in which the diagnosis was difficult.

Case Report

A seven-year-old girl presented exophytic tumors about three cm in diameter in both inguinal areas (Figure 1). The skin was verrucous and hyperkeratotic, and the lesion looked like a wart. A nodule with a small black dot was observed in the center of the mass. The mass was not painful and the inguinal lymph nodes were

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not enlarged. The surrounding skin showed erythema and hyperthermia. According to the patient, the tumor had developed three months before. She repeatedly had presented similar lesions in the inguinal area in the past, however the girl had hidden the pathology from her mother, fearing manipulation with a needle. The patient presented more than a dozen typical tungiasis lesions on both feet.

As ectopic tungiasis was suspected, the tumor was excised on the right side, whereas on the left side 5% topical thiabendazole was applied. Two weeks later, the patient was seen again. The biopsied area had healed without complication and the tumorous mass on the contralateral side had disappeared completely. Microscopic examination showed a completely preserved vital and gravid *T. penetrans* female embedded in the skin (Figure 2). The epidermis was completely breached, so that the parasite was in direct contact with the dermis (head), as well as the outside environment (dorsum). The epidermis was markedly hyperplastic and had a pseudoepitheliomatous appearance, with extreme thickening of the rete Malpighii, causing a highly irregular, verrucous aspect of the skin. There was moderate thickening of the keratin layer, with a few Gram-positive cocci in the most superficial layers. There was an increased density of blood vessels in the dermis. T-cells were seen in moderate numbers near blood vessels and the parasite, together with smaller numbers of histiocytes and large numbers of neutrophils and eosinophils, in the upper part of the dermis. B-cells were present in a few small follicles. Here and there, a few mast cells could be seen. Spongiosis and incontinentia pigmenti, with phagocytosis of the melanin by histiocytes, indicated that the patient had repeatedly scratched the lesion.

Discussion

Diagnosis of tungiasis is easy when lesions are confined to the feet. This parasitic skin disease typically presents as an itching reddish-brown spot, with a diameter of one to two mm (early stage), a circular lesion presenting as a white patch with a diameter of 3-10 mm, with a central black dot (mature stage), a black crust surrounded by necrotic tissue (late stage with dead parasite), or as a lesion altered through manipulation by the patient (partly or totally removed sand flea, leaving a characteristic sore in the skin, or a suppurative lesion, caused by the use of non-sterile perforating instruments) [12].

Ectopic tungiasis lesions occur mostly on the hands, but they have been reported from various topographic sites. In an impoverished community in Northeast Brazil, 6% of tungiasis patients had lesions at topographic localizations other than the feet [13]. In these cases, diagnosis is more difficult, particularly when the macroscopic appearance is unusual, as in our patient.

The differential diagnosis of ectopic tungiasis includes various parasitic infestations, such as myiasis, bites by *Pulex irritans* or ticks, scabies, cutaneous larva migrans and, in Africa, dracontiasis. *Tunga penetrans* can also mimic deep mycosis, verrucae, foreign bodies or narcotizing vasculitis [14,15]. Atypical tungiasis has to be differentiated from pseudoepithelioma, condylomata acuminata and cutaneous tuberculosis. In these cases, histopathological examination will yield the correct diagnosis as the parasite is easily identified [15]. If the parasite has already disintegrated or sloughed from the skin in late stage lesions, the histopathological picture may be mistaken for any infection causing pseudoepitheliomatous hyperplasia, e.g. chromoblastomycosis or cutaneous leishmaniasis.

Interestingly, the patient reported that the lesions had first appeared three months previously. As sand fleas die *in situ* once all eggs have been expelled, i.e. up to three weeks after penetration, and since the parasite was viable in the biopsied lesion, continuous re-infection probably took place at this topographic site. In fact, free-running females seem to prefer to penetrate close to already-existing lesions. This may explain why the lesions persisted in the inguinal areas, eventually leading to pseudoepitheliomatous hyperplasia. When interviewed, the girl admitted that she liked to sit playing undressed in the sand in front of her home, where she likely became repeatedly infected.

Figure 1. Ectopic tungiasis in both inguinal areas of a seven-year-old girl, presenting as exophytic tumors. Expelled eggs are visible on the skin of the thighs.

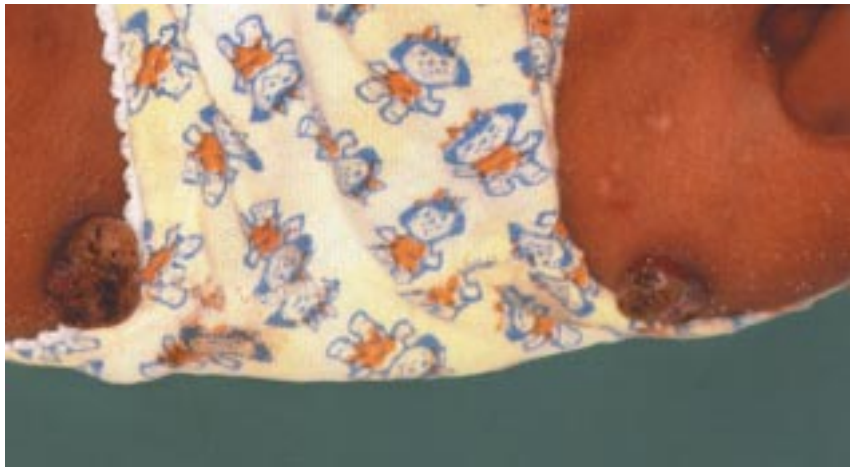
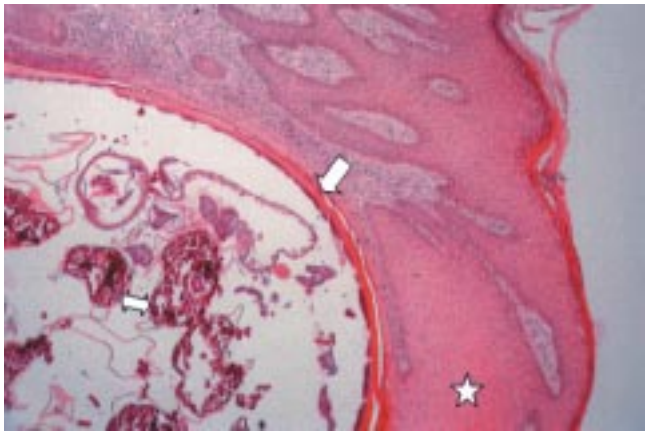


Figure 2. Segment of the parasite, embedded in the corneal layer of the epidermis. The epidermis (star) shows pronounced hyperplasia. The cuticle of the parasite is marked (arrow) as well as some eggs (double arrows).



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