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Case number: 4091/21 Material distributed: 1 glass slide

Title of Case Presentation Leishmania-induced rapidly growing eyelid tumor

Clinical History:

A 65-year-old woman presented with a rapidly growing tumor in the left upper eyelid. She has no medical history of interest, she is immunocompetent and blood tests show parameters within normal limits with complete blood count and sedimentation rate.

The clinical suspicion is of invasive neoplasia, most probably basal cell carcinoma, so surgery with complete resection of the lesion is proposed.

Ophthalmic Pathology

The excised lesion measured 2.3 x 1.8 cm and was marked with two suture threads on the upper border and one suture thread on the lower border. The lesion appeared exophytic with a central umbilication.

The microscopic study revealed a thick superficial stratum corneum with necrotic scar, collections of polynuclear cells and a central invagination with large areas of superficial epidermal necrosis. Deep epidermal growth was seen with elongated ridges and cords penetrating deep into dermal stroma. Some squamoid eddies were evident, but no horn pearls. However, what was most prominent was the intense lymphoid infiltrate with a diffuse distribution. Apart from the polynuclear neutrophils located on the surface, in mid and deep dermis an infiltrate of lymphocytes and histiocytes with numerous mature-appearing plasma cells was observed. In some areas there was a configuration of granulomatous structures, with epithelioid histiocytes but without central necrosis and without Langhans-type giant cells. Some mitoses were observed among the lymphoid cellularity but not in the epithelial component, which did not show cell atypia or invasive growth, but rather a pseudoepitheliomatous hyperplasia. Studying the case, at high magnification, very small punctiform structures between 1 and 2 microns were visible inside the cytoplasm of the macrophage cells. Some appeared with dual morphology and characteristics compatible with Leishmania.

An immunositochemical study was performed that showed positivity in intra-macrophage parasitic structures with monoclonal antibodies for Leishmania (clone gp63 ThermoFisher). Expression for CD1a was also found, in a cross reaction that this antibody presents for Langerhans cells in Leishmania amastigotes [1].

Lymphoid cellularity had a heterogeneous pattern with a predominance of T lymphocytes (cd3+) and a significant proportion of B lymphocytes (CD20+). Plasma cells expressed a polyclonal pattern (kappa +, lambda +).

A diagnosis of cutaneous palpebral leishmaniasis was issued, and subsequently a paraffin-embedded sample was sent to an external laboratory, which allowed Leishmania DNA to be amplified with PCR.

Discussion:

There are three main forms of leishmaniasis: visceral, cutaneous, and mucocutaneous.

It is caused by protozoa of the Leishmania genus transmitted by the bite of infected female sandflies (Phlebotomus mosquitoes and Lutzomya-type flies), which parasitize the cells of the reticuloendothelial system [2].

It affects the poorest populations on the planet and it is estimated that new cases per year range between 700,000 and one million. Only a small part of people infected by parasites that cause leishmaniasis end up suffering from the disease.

Cutaneous leishmaniasis is the most frequent form and produces skin lesions on exposed areas of the body, especially ulcers, which leave scars. Most of it is recorded in the Americas [3], the Mediterranean basin, the Middle East and Estearn Asia [4]. Specifically, the eastern Mediterranean basin is where 70% of the world's cases of cutaneous leishmaniasis occur.

The clinical presentation of cutaneous leishmaniasis is characterized by its great polymorphism [5], atypical forms or unusual sites [6].

These forms have been described as: lupoids, sporithrocoids, eczematiformes, verrucous, zosteriformes, erysipeloid, eyelids, squamous cell carcinoma type...

Among these atypical or unusual forms of presentation, the involvement of the eyelids is exceptional and is a difficult diagnosis in routine medical practice [7]. Although molecular studies are available [8, 9] a clinical suspicion is need to order appropriate test.

In most cases, eyelid leishmaniasis is a disease limited to the skin, but exceptionally it can affect other structures of the eye with serious ocular complications: conjunctivitis [10], ectropion [11] keratitis, and uveitis. In large series of cutaneous leishmaniasis, its prevalence is estimated between 0.27 and 0.72%.

The rarity of this location, despite being an exposed skin area, could be explained both by the protection of the eyelashes and by the continuous movements of the eyelids, which prevent the bite of the vector and the infection of the skin in this region.

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