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An itching plaque

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The patient

A 30-year-old woman presented to our hospital in Manacor, Mallorca, with a new lesion on the thorax that had appeared three to four months earlier, had gradually enlarged and was slightly itchy. The patient was otherwise healthy. Physical examination revealed a tender, brownish papule or plaque contacting the elastic band of the underwear under the right breast (Figure 1). The remaining physical exam showed no other relevant lesions.

Dermoscopic examination showed an ill-defined red-yellowish background with a small, pigmented, structureless area. Tiny, dotted and comma vessels were barely visible. Remarkable were numerous rounded or droplet-shaped, whitish structures spread all over the lesion (Figure 2).

A 3 mm punch biopsy was obtained showing epidermal acanthosis and an intense superficial dermal inflammatory infiltrate. The infiltrate was composed by neutrophils, eosinophils and plasma cells, intermingled with bigger epithelioid cells, some of them containing tiny, round, basophilic structures in their cytoplasm (Figure 3).

What is your diagnosis?

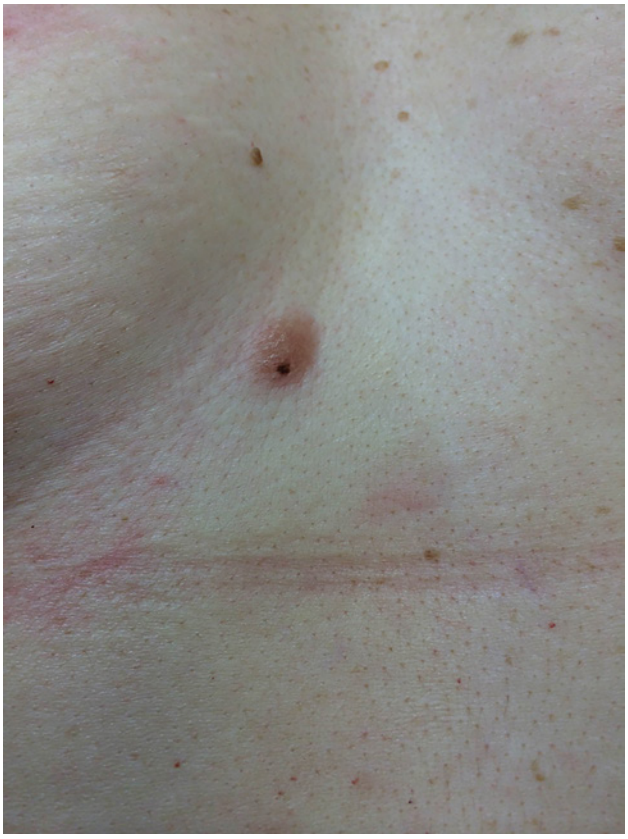

Figure 1. A brownish papule or plaque contacting the elastic band of the underwear under the right breast. (Copyright: ©2015 Garcias-Ladaria et al.)

Answer

Cutaneous leishmaniasis

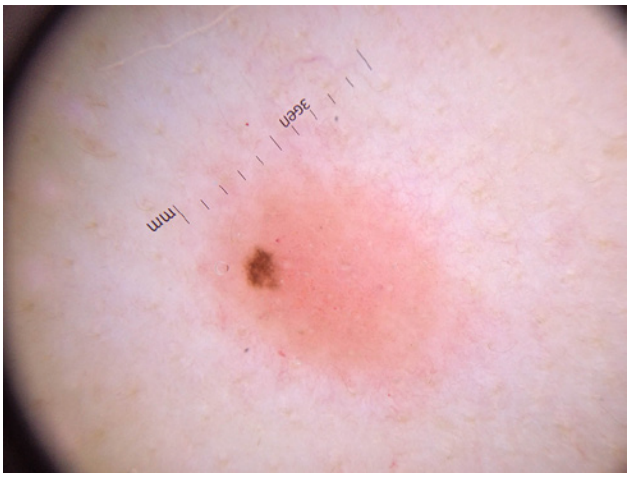

Figure 2. Numerous rounded or droplet-shaped, whitish structures spread all over the lesion. (Copyright: ©2015 Garcias-Ladaria et al.)

Clinical course

The patient was scheduled to receive intralesional antimonials.

Answer and explanation

Leishmaniasis is an infectious disease caused by several species of the genus *Leishmania* trasmitted by sandflies of the genera *Phlebotomus* and *Lutzomiya*. It is a major health problem, endemic in more than 70 countries worldwide, probably widely underreported, with estimated global annual incidence of 0.2 to 0.4 million cases for visceral leishmaniasis (VL), and 0.7 to 1.2 million cutaneous leishmaniasis (CL) [1]. While 90% of VL concentrate in six countries—India, Bangladesh, Sudan, South Sudan, Brazil and Ethiopia, CL is widely distributed in the Americas, the Mediterranean basin, and western Asia from the Middle East to Central Asia. Spain is a hypoendemic country with 0.41 new cases per 100,000 inhabitants annually [1]. The disease is specially diagnosed on the Mediterranean basin, but a recent flare of the disease around Madrid, far from the coast, has raised concern and shows how Leishmaniasis is still nowadays an important issue in public health and is far from eradication [2,3].

Leishmaniasis is generally regarded as a zoonotic infection, although antroponotic contagion has been also reported. Reservoirs are mammal hosts (mainly marsupials, rodents, edentates, and carnivores). More than 20 species of *Leishmania* are infectious to the human being. They are in turn transmitted by more than 30 species or subspecies of sandflies. Vector-related factors, like their saliva, have been shown to contribute to the viability and morphology of the infection [3]. Additionally, host factors like an effective Th1 response has been shown to be capital in controlling and destructing infection on the skin [3]. Immunosuppression, particularly HIV co-infection, has been regarded as a matter of concern. HIV and *Leishmania* can both share the transmitting source (basically contaminated needles) and are synergistic pathogens.

CL can be divided in disseminated CL, localized CL and mucous leishmaniasis. While disseminated and mucous CL, like VL, are life-threatening conditions, localized CL tends to self-resolution. Still, it can lead to permanent scarring and disfigurement, especially in cosmetic areas, and correct diagnosis and early treatment is paramount. Combined with variability in host response, parasitic and vector factors result in a myriad of skin lesions that can be challenging, even for experienced clinicians. The typical lesion starts with erythema at the site of a bite of an infected sand-fly. The erythema develops into a papule, then a nodule that progressively ulcerates in a variable time frame raging from weeks to months. They are usually found in exposed areas, especially the face and arms and can be solitary or multiple. Atypical presentations are hyperkeratotic, verrucous or papillomatous papules or plaques, with eczematiform, zosteriform, erysipeloid or sporotrichoid configurations. The skin lesions can also simulate connecting tissue diseases (lupus erythematosus, dermatomyositis) or tumors [4,5].

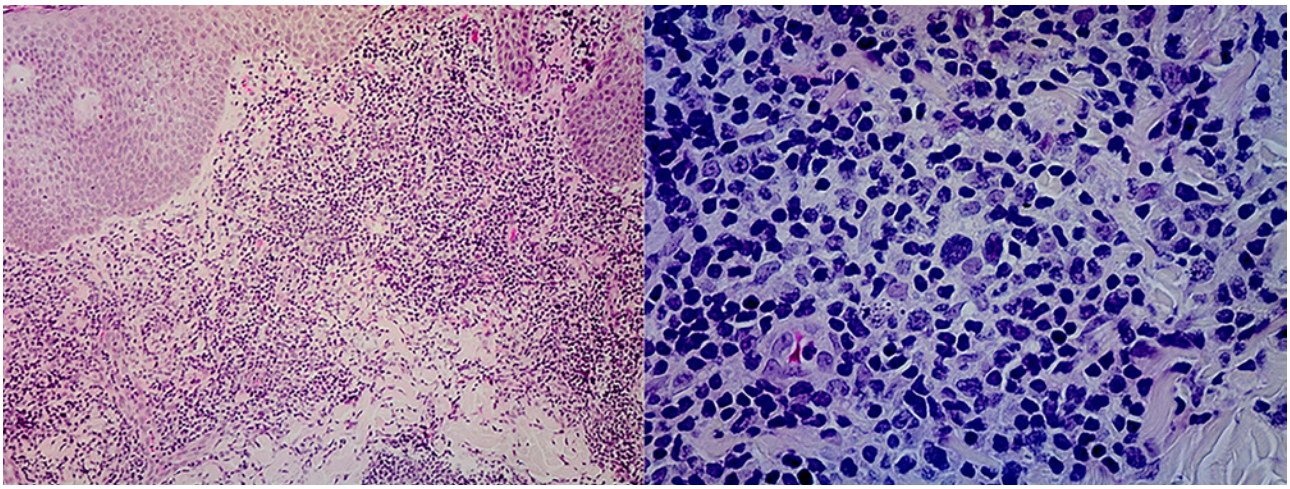

Figure 3. Epidermal acanthosis and an intense superficial dermal inflammatory infiltrate. (Copyright: ©2015 Garcias-Ladaria et al.)

As in other inflammatory and infectious diseases [6,7], dermoscopy has emerged as a valuable tool for the bedside diagnosis of cutaneous leishmaniasis. In 2008, a Spanish group described the dermoscopic criteria of cutaneous leishmaniasis for the first time [8]. They described four patterns. Pattern 1 (early lesions) more often showed vascular structures (comma-shaped vessels, linear irregular vessels and polymorphous /atypical vessels) and "yellow tears," while pattern 2 (later lesions) displayed a central erosion/ulceration and hyperkeratosis surrounded by "white starburst-like pattern" and vascular structures on the periphery. Pattern 3 was a combination of patterns 1 and 2, and pattern 4 consisted of vascular structures alone. Vascular morphology appeared to be varied: comma-shaped vessels, linear irregular vessels, dotted vessels, polymorphous/atypical vessels, hairpin vessels, arborizing telangiectasia, corkscrew vessels, and glomerular vessels are described. Eighty-eight percent of the lesions had two or more vascular patterns and no specific pattern or arrangement was found.

More recently changes in reflectance confocal microscopy (RCM) of CL have been described [9], namely linear and comma-shaped vessels, follicular plugging, presence of multinucleated giant cells and mixed inflammatory infiltrate, and the more specific "brick-like structures" which are bright polygonal structures, not described elsewhere.

However, diagnosis confirmation still relies on visualization of the parasite under the microscope by culture, smears or biopsy [3]. Biopsy samples show a polymorphous infiltrate with plasma cells and variable epidermal response (acanthosis/atrophy/ulceration). While late lesions show well organized, tuberculoid granulomas, early lesions show spread macrophages intermingled within the infiltrate, containing amastigotes, the so-called Leishman-Donovan bodies. To ensure that the visualized structures are amastigotes an experienced observer should look for the characteristic size (2–4 μm), shape (round to oval), and internal organelles (the nucleus and kinetoplast). CL is frequently misdiagnosed in countries where it is not endemic, particularly if organisms are not seen [10]. Histological differential diagnoses are sarcoidosis, foreign body reaction, granulomatous rosacea and granuloma annulare.

Cultures show the growth of promastigotes, the flagellated infective form, found in vectors. Polymerase chain reaction (PCR) can be performed on biopsy samples with high specificity. Serology is not generally performed in CL.

First-line treatment, according to the WHO, are parenteral pentavalent antimonial drugs at 20 mg/kg per day for 20-28 days. However, for localized CL, periodic intralesional injections every four weeks are widely accepted as the standard treatment and achieve high rates of cure [3]. Other treatment options include other antibiotics, physical therapies (cryotherapy, thermotherapy, Co2 laser therapy), surgery and imiquimod.

In conclusion, we present a case of localized cutaneous leishmaniasis in an atypical location. Constant rubbing of the underwear gave the lesion the appearance of an irritated tumor, in our view easy to misdiagnose as an irritated melanocytic nevus or an irritated seborrheic keratosis. Dermoscopy was determinant in the decision to take a biopsy. Leishmaniasis is a common disease in extensive parts of the world, currently far from epidemiological control, and dermatologists and dermatopathologists should be familiar with it.

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