Multifocal cutaneous leishmaniasis

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Summary

Cutaneous leishmaniasis is an infectious disease that is caused by protozoa belonging to the genus Leishmania and typically manifests with a single lesion on sun exposed areas. It is frequent in the Mediterranean Basin where it is most commonly due to Leishmania Infantum. In the literature, there is a number of reports describing atypical presentations, including that manifesting with multiple lesions or those mimicking other conditions, such as infections, tumours and inflammatory diseases. Here, we describe a case of cutaneous leishmaniasis with two unusual aspects: the multifocal presentation and the fact that it resembled sarcoidosis, a systemic granulomatous disease whose cutaneous picture is also polymorphous.

KEY WORDS: Cutaneous leishmaniasis; Leishmania infantum; sarcoidosis.

Introduction

Leishmaniasis is an infectious disease, diffused worldwide with the exception of Australia, the Pacific Island and Antarctica. It is caused by a protozoa of the genus *Leishmania* (*L.*) that is transmitted by the bite of a sandfly that, in the Old World, belongs to the genus *Phlebotomus* (*P.*) sp.. Reservoirs are represented by dogs, mice, rats, wild rodents and, more rarely, humans. Cutaneous leishmaniasis (CL) is very frequent in the Mediterranean Basin where is usually caused by *L. infantum*, transmitted by *P. perniciosus* and *P. perfiliewi*, and it manifests with a single, polymorphous lesion on uncovered areas, mainly the face (1-6). We describe the case of a female patient with an atypical, multifocal presentation of CL.

Case report

A 68-year-old woman, from Calabria (Southern Italy), was admitted to our Department because of some papular-nodular lesions located on her left shoulder and arm. The patient stated that she was in good general health and that she was not under treatment with systemic drugs. The patient also stated that the skin lesions appeared some months earlier and that they were asymptomatic. Dermatological examination revealed the presence of eight papular-nodular lesions, of different morphology and size (3 to 10 mm), red in colour, with a smooth surface, of parenchymatous-hard consistency. Four lesions were located on the left shoulder and four on the left arm (Figure 1A). A clinical diagnosis of cutaneous sarcoidosis was made. General physical examination was normal. Laboratory examinations included complete blood count, protein electrophoresis, renal tests (urea, creatinine and electrolytes), hepatic tests (bilirubin, transaminases, γ-glutamyltransferase, alkaline phosphatase, cholinesterase, lacticodehydrogenase), inflammatory tests (erythrocyte sedimentation rate, mucoproteins, C-reactive protein), angiotensin-converting enzyme (ACE) and urinalysis: all examinations were within normal ranges. Instrumental investigations (chest X-ray and liver ultrasonography) were negative. A skin biopsy was performed. Histopathological examination showed a dense inflammatory infiltrate in the superficial and mid dermis, mainly consisting of histiocytes, lymphocytes and plasma cells (Figure 1B). A histopathological diagnosis of CL was hypothesized. Culture on Novy-MacNeal-Nicolle (NNN) medium was positive for *Leishmania* sp. Molecular biology study, by means of polymerase chain reaction amplification, demonstrated the presence of gene sequences of *Leishmania infantum*. A diagnosis of multifocal CL was made. The patient was successfully treated with cryotherapy. Follow up (8 months) was negative.

Discussion

The clinical picture of CL is polymorphous, ranging from papular to nodular to ulcerated lesions (2). There is a

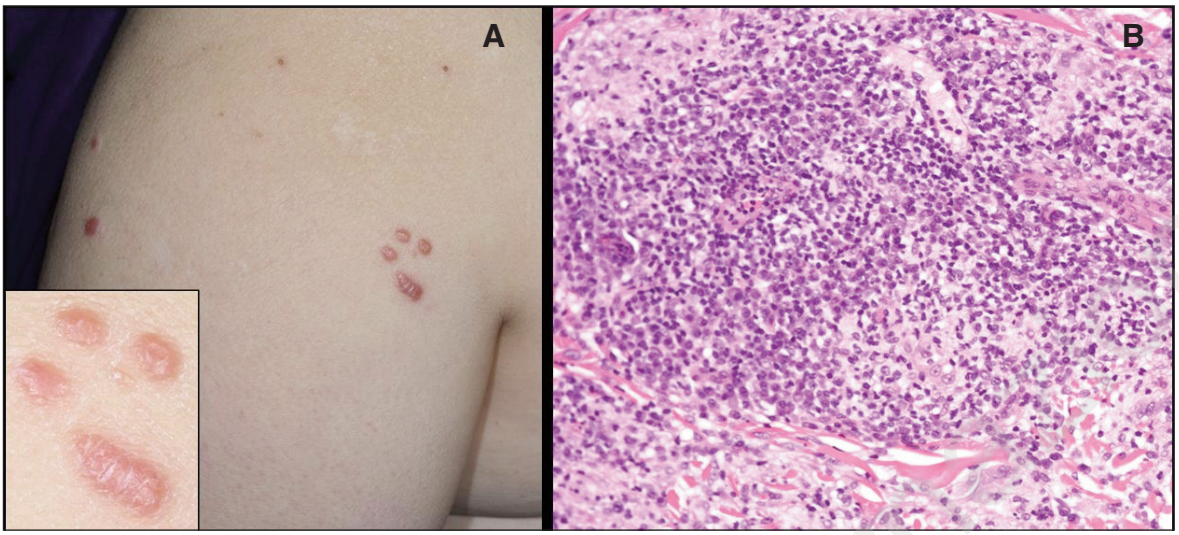

Figure 1 A, B - A) Erythematous papular-nodular lesions on the patient's left arm. Insert: high-power view of the lesions. B) Skin biopsy specimens showing a dense dermal inflammatory infiltrate, mainly consisting of histiocytes, lymphocytes and plasma cells (Hematoxylin-Eosin stain; original magnification, x 200).

number of reports describing CL mimicking different skin diseases, including infections as well as inflammatory and neoplastic conditions (3-8). The clinical presentation of CL in the patient we have just described was reminiscent of sarcoidosis, an inflammatory, systemic disease whose cutaneous picture is also polymorphous (9). To our knowledge, only two cases of CL clinically resembling sarcoidosis have been reported so far: one of which was characterized by multiple skin lesions (10). On the other hand, some cases of CL localized to the head, particularly the nose, eyelids and ears, that mimicked a number of granulomatous diseases, have been reported (11-16), suggesting to consider CL in the differential diagnosis of granulomatous diseases occurring in sun exposed areas. Moreover, patients with sarcoidosis have an increased risk for infections, such as leishmaniasis, due to both the corticosteroid therapy and the disease in itself (17). Our case was interesting also because it was characterized by multiple skin lesions. In fact, CL usually presents with a single lesion in the site of the vector sting (2). Paradisi et al. first described "multifocal CL", consisting of multiple, slightly inflammatory, papules and nodules, on exposed areas (18). Subsequently, Maniscalco et al. reported 29 cases of this rare clinical presentation observed over a period of five years; all of them were caused by *L. infantum* (1). Multifocal CL might be caused by species of sandflies which are different from *P. perniciosus* and *P. perfiliewi*. It is known that these sandflies are lonely arthropods which do not live in swarms. Furthermore, sandflies usually sting just once for their feeding. It is therefore possible that multifocal CL is caused by sandflies which live in swarms (e.g. multiple lesions are the clinical results of multiple stings caused by several sandflies) or by a single sandfly which stings several times. No new pathogenetic hypotheses for multifocal CL have been proposed so far. It is of note that our patient was successfully treated with cryotherapy. The latter was chosen because it is, in our clinical experience, effective and safe in approximately 85% of patients (unpublished data). In the past we also used N-methylglucamine antimonate, both by intralesional and intramuscular route; however, it is anymore on the Italian market. In conclusion, we described a case of multifocal CL mimicking sarcoidosis to emphasize the fact that its different clinical presentations may lead to clinical misdiagnosis.

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