

Lower Lip Leishmaniasis Mimicking Squamous Cell Carcinoma

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Abstract

With a global prevalence estimated at 0.05-0.27% of all cutaneous leishmaniasis (CL), and 2.7-4.9% of atypical clinical presentation of CL, lip leishmaniasis is one of the rarest unusual presentations of this disease. Its classically reported in immunocompromised patient, and represents a real diagnostic challenge for clinicians.

We present an original case of lower lip leishmaniasis mimicking squamous cell carcinoma in 26-years-old immunocompetent Tunisian man.

As rare as it is, this atypical clinical presentation of CL deserves to be well known by clinicians, and should be considered in the differential diagnosis of lip lesions in patient living or traveling in Leishmania endemic area.

Keywords: zosteriform leishmaniasis, Cutaneous leishmaniasis, Lip, Atypical presentation, Leishmaniasis.

INTRODUCTION

Cutaneous leishmaniasis (CL) is a common and neglected worldwide skin disease [1], characterized by diverse clinical presentations (atypical shape or unusual site) [2-4]. It is therefore described as intriguing and multifaceted disease [3]. These unusual forms (2-5% of all CL) [2,3,5] are classically reported in immunocompromised patient and represent a real diagnostic challenge in current medical practice [2-5].

Lip leishmaniasis is exceptional with only few sporadic cases in the world literature [3,4,6].

We present an original case of lower lip leishmaniasis mimicking squamous cell carcinoma in a young immunocompetent Tunisian man.

CASE REPORT

A 26-years-old Tunisian man, without medical history, was referred to our consultation for suspicion of squamous cell carcinoma of the lower lip.

The diagnosis was suspected in front of chronic, dragging, painless, and ulcerative lesion of the lower lip evolving for 6 months.

The somatic examination noted an ulcero-crusted nodular lesion of the right lower lip (Fig. 1), measuring

3 cm from the long axis, and isolated without any other associated cutaneous or mucosal lesions (Fig. 2).



Figure 1. Ulcero-crusted nodular lesion of the right lower lip.

Basic biological tests were within normal limits, as well as chest X-ray and dental panoramic X-ray.

The interrogation revealed that the patient was recently affected in an endemic region for cutaneous leishmaniasis in southern Tunisia.

Diagnosis of CL was confirmed by demonstration of Leishmania amastigotes in Giemsa-stained smears of the lesion, and polymerase chain reaction identified Leishmania infantum.

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Figure 2. *Isolated lower lip involvement without other cutaneous or mucosal lesion*

The patient was treated with intralesional injections of meglumineantimoniate for 10 days, with rapid and favorable evolution.

DISCUSSION

Lip leishmaniasis is very unusual with only few sporadic cases reported in the medical literature [2,4,6]. Indeed only two patients were noted in Bari AU et al series of 718 patients with CL (0.27% of all CL and 4.9% of atypical CL) [3], only one patient was noted in Raja KM et al series of 1709 patients with CL (0.05% of all CL and 2.7% of unusual CL) [2], and in the largest series of CL of Yesilova Y et al, prevalence of lip location was only 4.3% from 14,400 patients [7]. It is classically reported in immunocompromised patient [4], but seems to be an emerging clinical presentation of CL in endemic [8] and even in non-endemic countries of this disease [6].

Clinical pattern of lip leishmaniasis include chronic ulcer, nodular and crusted lesions, lip erysipeloid inflammation, and cheilitis [2,4,6,8-11]. The lesion can affect both the upper and lower lip, be single or multiple, and be isolated or associated with other facial lesions of cutaneous leishmaniasis [4,7,9,10]. This great polymorphism of this localization explains that it is most often overlooked with a significant diagnostic delay [8-11]. Indeed, the mean diagnosis delay in Hammami-Ghorbel H et al series was 6.9 months [8].

Differential diagnoses of lip leishmaniasis may include other infectious diseases like fungal infections or cutaneous tuberculosis, granulomatous cheilitis, Melkersson-Rosenthal syndrome,

orofacial granulomatosis, Wegener granulomatosis, sarcoidosis, squamous cell carcinoma, basal cell carcinoma, and foreign body reaction [4,9,10].

Lip leishmaniasis can be successfully treated with systemic or intralesional injections of meglumineantimoniate and its prognosis is usually favorable [7,10].

CONCLUSION

Lip leishmaniasis is very rare and very unusual presentation of cutaneous leishmaniasis. It deserves to be known by any clinician to avoid delayed diagnosis. This diagnosis should, thereby be considered in the differential diagnosis of lip lesions in patient living or traveling in Leishmania endemic area.

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