RESEARCH ARTICLE

Leishmaniasis of the Lip : Report of a Case

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Summary
Leishmaniasis is a parasitic disease caused by a protozoan flagellate of the genus Leishmania.
This parasite infects numerous mammal species including humans.
It is transmitted through the infective bite of a sand fly called phlebotominae.
Cutaneous Leishmaniasis is the most common form of Leishmaniasis.
We present the case of a 47 years- old man who came to our Department of oral and maxillo-facial surgery, complaining about a painless lesion on his lower lip.
After removal of the lesion and microscopic examination, the diagnosis of Leishmaniasis was made.
This clinical case illustrates that the differential diagnosis of a nodular painless lesion of the lip must include rare diagnosis like a cutaneous Leishmaniasis, especially in patient from a geographic origin which represents an area at risk for Leishmaniasis.

Résumé
La Leishmaniose est une maladie parasitaire causée par un protozoaire flagellé du genre Leishmania.
Ce parasite infecte de nombreux mammifères, dont l'homme.
Il se transmet par la piqûre infectante d'une mouche appelée phlébotome.
La forme cutanée est la forme la plus fréquente de Leishmaniose.
Nous présentons le cas d'un patient âgé de 47 ans, qui s'est présenté à notre Département de Stomatologie et Chirurgie Maxillo-faciale pour une lésion non douloureuse de sa lèvre inférieure.
Après examen anatomo-pathologique de la pièce de résection, le diagnostic de Leishmaniose cutanée de la lèvre fut posé.
Ce cas clinique illustre que le diagnostic différentiel d'une lésion nodulaire non douloureuse de la lèvre doit inclure des diagnostics rares tels qu'une Leishmaniose cutanée, chez les patients dont l'origine géographique représente une région à risque pour cette maladie.

Case report
A 47 years- old man came to our Oral and Maxillo-facial Surgery Department complaining about a lesion on his lower lip.
He reported that the lesion appeared about 7 months before.
The lesion was painless and the patient had no complain except esthetic concern.

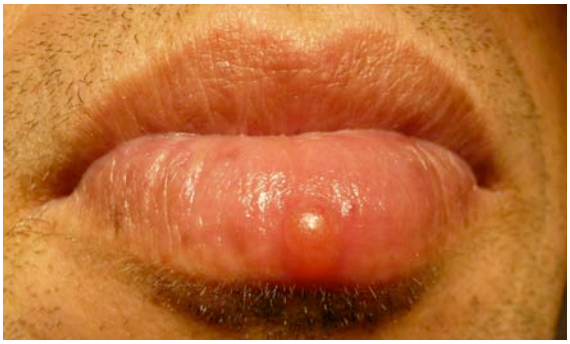

Figure 1. Nodular lesion of the cutaneous border of the lower lip

From his past history, we noticed that he traveled to Morocco each summer to visit his family. He had no particular surgical or medical background.
The clinical examination revealed a round nodular lesion in the middle of his lower lip, measuring 6mm x 5mm.

The lesion was slightly shiny and barely pink colored.
Palpation of the lesion revealed a soft consistency and was totally painless.
No adenopathy was associated, no fever neither.
A blood test was performed and did not show any abnormality (Figure 2).

Blood Test

Numération		
Leucocytes	6.1	x10^3/mm³
Globules rouges	5.52	x10^6/mm³
Hémoglobine	14.7	g/dL
Hématocrite	44.8	%
MCV	81.2	µm
MCH	26.7	pg
MCHC	32.9	g/dL
RDW	11.8	%
Plaquettes	253	x10^3/mm³
MPV	9.05	µm³
PCT	0.229	%
PDW	15.9	
Viabilité	0.974	
Formule leucytaire		
Polyneutrophiles	47.5	%
Polyneutrophiles absolus	2.88	x10^3/mm³
Lymphocytes	39.1	%
Lymphocytes absolus	2.37	x10^3/mm³
Monocytes	9.1	%
Monocytes absolus	0.553	x10^3/mm³
Eosinophiles	3.2	%
Eosinophiles absolus	0.196	x10^3/mm³
Basophiles	1.1	%
Basophiles absolus	0.069	x10^3/mm³
Marqueurs inflammatoires		
Vitesse de sédimentation	2	mm/H
CRP	0.088	mg/dL
Chimie		
Urée	* 52	mg/dL
Créatinine	0.8	mg/dL
Filtr. glomérul.(calcul-MDRD)	>90	mL/min/1.73 m²
Sodium	140	mmol/L
Potassium	4.0	mmol/L
Chlore	103	mmol/L
CO2 total	29	mmol/L
Trou anionique	12	mmol/L
Phosphatases alcalines	56	UI/L
G-glutamyl-transférase	34	UI/L
Ala. aminotransférase (GPT)	34	UI/L
Asp. aminotransférase (GOT)	21	UI/L
Lactate déshydrogénase	130	UI/L
Endocrinologie		
Glucose	* 133	mg/dL

Clinically, this round nodular painless lesion looked like a benign or other slow growing lesion.
The differential diagnosis of a nodular lesion of the lip included a lesion from epidermic origin: veru planum, pityriasis rubra, botryomycoma, squamous papilloma (1).
A dermic origin could have also been considered (oedema, purpuric vascularitis, dysmetabolic amylosis, benign tumor (secondary syphilis) (2, 3) or a neoplasia (basal cell carcinoma) (4), as well as a tumor from mesenchymal origin (fibroma, lipoma, and neuroma)
We decided to remove surgically the lesion through local anesthesia.
Microscopic examination of the specimen showed a dermal inflammatory cellular infiltrate, composed of lymphocytes, plasma cells

and numerous macrophages containing small ovoid corpuscles (Figure 3).

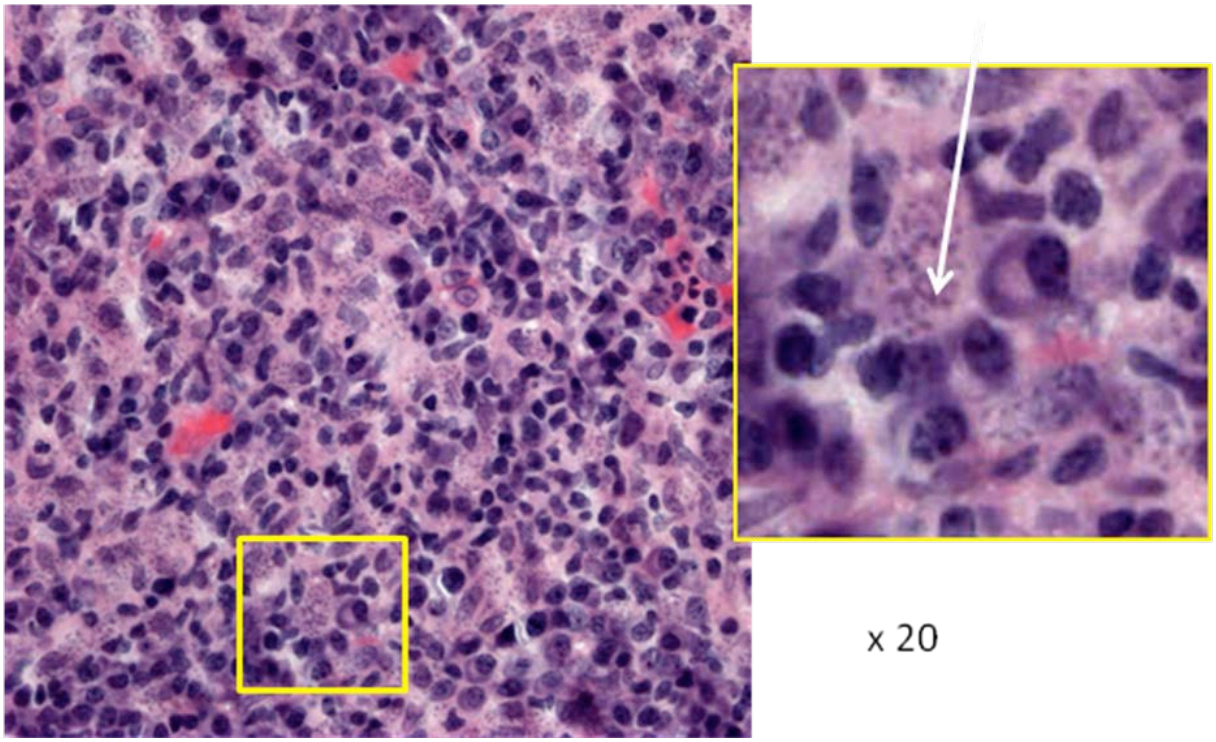

Figure 3: The microscopic examination (Hematoxylin-Eosin × 20) shows a dermal inflammatory infiltrate composed of plasma cells and lymphocytes, and macrophages containing small ovoid corpuscules (arrow)

These corpuscles appeared to be intracellular amastigotes, which are typical of Leishmaniasis disease.

The clinical examination, together with the anatomopathologic examination leads us to the diagnosis of cutaneous Leishmaniasis of the lower lip.

Discussion

Leishmaniasis is a parasitic disease transmitted through the bite of an insect vector, the phlebotomine sand fly.
According to the World Health Organization, 350 million people are at risk of contracting a Leishmaniasis in eighty-eight countries in 4 four continents, and 1.5 to 2 million new cases of Leishmaniasis occur each year around the world (5)

This disease is rare in our countries, but represents a major public health problem in numerous underdeveloped countries.

The bite of an infected sand fly results into intradermal inoculation of metacyclic promastigotes. These promastigotes are phagocyted by macrophages.
Inside these macrophages, they transform into amastigotes, multiply, and finally destroy the infected cells (6)
The localization of the parasite into various tissues result in different clinical manifestations: Either the localized cutaneous Leishmaniasis, at the bite site. The lesions are confined to the skin (7). The other type is the Muco-cutaneous Leishmaniasis which extends deeper into the mucosa and the cartilages (8). The third type is the diffuse cutaneous Leishmaniasis which extends to all the skin. This type is very rare and frequently associated with concomitant HIV infection (9). Finally, the last type is known as visceral Leishmaniasis. This one is life threatening, as the parasite infects the liver, the spleen and the bone marrow, resulting into hepato-splenomegaly, weakness and frequent secondary infections (10; 11)

Intervention strategies for control or prevention are made difficult by the fact that there are many different host animals, and a multiplicity of sand fly vectors.
The aim of prevention is to avoid host infec-

tion and its subsequent disease.
For example, ways to protect people against sand fly bite (consist in using special bed nets or wearing full clothes).
Control programs are generally limited, and forest removal itself has been proven to be unefficient. (12)

According to the different possible clinical manifestations of Leishmaniasis, different treatment regimens have to be considered, depending on the level of gravity.
The treatment of localized cutaneous Leishmaniasis depends on the type and characters of the lesion, and the patients conditions. Briefly, there are three options: Either therapeutic abstention, or a local treatment. This treatment consists in local administration of pentavalent antimonials (13), or a surgical removal of the lesion as we did in this present case , or finally, a physical therapy as cryotherapy, laser, or electrocoagulation (7). The systemic treatment is preferred in case of immunodeficiency or large and multiple lesions. It usually consists into intravenous injections of pentavalent antimonials (13).

The treatment of muco-cutaneous and diffuse Leishmania consists as well in systemic intravenous treatment (8, 9). Finally, the treatment of visceral Leishmaniasis needs in conjunction a correction of nutritional deficiencies in one hand, and in the treatment of secondary infections in the other hand and blood supply if needed (10, 11).

Conclusion

This clinical case illustrates that the differential diagnosis of a nodular painless lesion of the lower lip can also include rare lesions like a cutaneous Leishmaniasis, especially in patient from a geographic origin which represents an area at risk for Leishmaniasis.

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