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Oral manifestations as indicators of visceral leishmaniasis progression : Case report

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Abstract :

Introduction :

Leishmaniasis is a group of neglected tropical diseases caused by protozoa of the genus Leishmania. It is a vector-borne disease transmitted by the bite of insects known as sandflies. Leishmaniasis is classified as cutaneous, mucocuta neous, and visceral or kala-azar.

Case report :

A 50-year-old type 2 diabetic patient with visceral Leishmaniasis (VL), followed at the infectious diseases department of the Fattouma Bourguiba University Hospital, Monastir, Tunisia was referred to the department of dental medecin for biopsy of an ulcerative painful lesion on the anterior part of the palate.

Conclusion :

The dentist plays a crucial role in the diagnosis and management of oral manifestations of VL.

Keywords : visceral leishmaniasis, oral manifestations, biopsy, treatment, amphotericin B

Introduction :

Leishmaniasis is a chronic disease caused by infection with a protozoan of the genus Leishmania. It is a vector-borne disease transmitted by the bite of infected sandflies.

Leishmaniasis is classified as cutaneous, muco-cutaneous, and visceral (VL)or kala-azar. The latter is considered the most serious form of leishmaniasis.(1)

It affects internal organs such as the spleen, liver and bone marrow. Although its manifestations are predominantly systemic(fever, weight loss, anemia, hepatomegaly, splenomegaly, anorexia and diarrhea), oral manifestations can occur, albeit rarely. These manifestations are more frequent in immunocompromised patients.

Case report :

A 50-year-old type 2 diabetic patient with VL, followed at the infectious diseases department of the Fattouma Bourguiba University Hospital, Monastir, Tunisia was referred to the department of dental medecin for biopsy of an ulcerative lesion on the anterior part of the palate that had been evolving for 17 days, that was painful to palpation and caused discomfort when eating.

On examination, the maxilla was totally edentulous (figure1), the mandible was subtotally edentulous (persistence of the 33,35,36) (figure2) and there was an ulceration on the anterior part of the palate on the left side 2 cm in diameter with an irregular contour, covered with a yellowish coating (figure1).

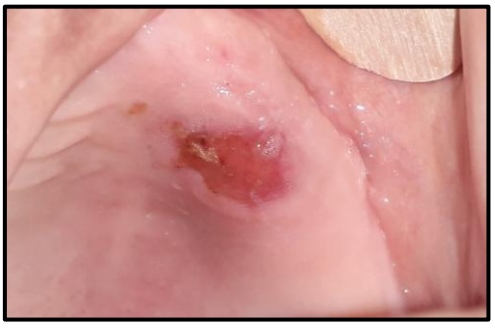

Figure1 : maxillary arch

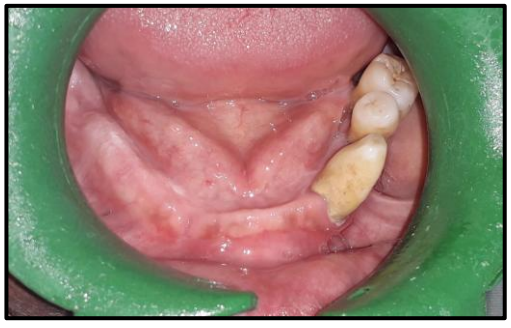

Figure2 : mandibular arch

Laboratory tests revealed pancytopenia (anemia 7.9g/dL, leukopenia 2700/mm3, and thrombocytopenia 29000/mm3) and elevated C-reactive protein (100mg/L).

Infectious blood serologies, including human immunodeficiency virus (HIV), hepatitis B and hepatitis C, were all negative.

Given the risk of bleeding, the biopsy was postponed and a platelet transfusion was indicated. Even after transfusion, the platelet count remained low (34,000/mm3), so the biopsy was contraindicated. The lesion was scraped and punctured with a fine needle for culture and PCR examination. The patient was also put on Liposomal amphotericin B by the infectious diseases department.

Then regular contol sessions were performed to assess lesion healing. (Figure2,3)

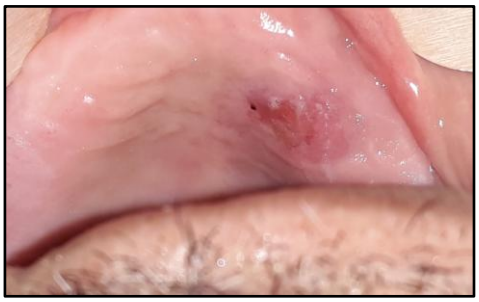

Figure2 : Check after 2 weeks showing beginning of healing

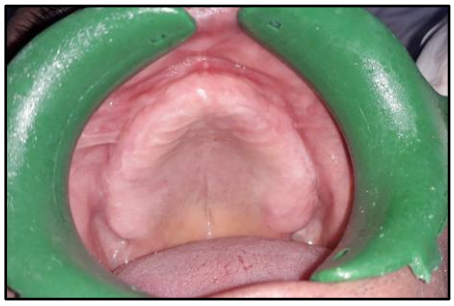

Figure3 : Check after 1 month showing complete healing of the lesion

After 2 months a normalization of the platelet value. So the residual teeth were extracted and the patient was referred for total bimaxillary prosthetic rehabilitation. (Figure4)

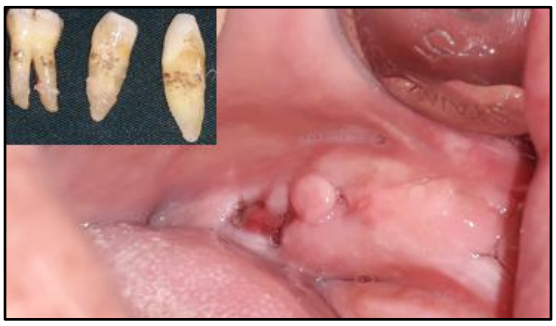

Figure4 : Extraction site for residual mandibular teeth

Discussion :

VL is a systemic disease caused by unicellular protozoans from the Leishmania genus, typically transmitted by female phlebotomine sandflies. Several species of Leishmania are known to cause the visceral form of the disease, of which L.donovani and L.infantum are the most common. The incubation period can vary between 2 weeks and 18 months, but the disease can take years to become symptomatic.(2)

Depending on the mode of transmission, two types of VL can be described: the zoonotic form, with dogs as the main reservoir, is found in the Mediterranean basin, China, the Middle East and South America. This form is caused by L infantum and mainly affects children and immunocompromised individuals.

Secondly, the antroponotic form, with human-to-human transmission and no animal reservoir, is much more frequent. It is caused by L donovani and is widespread in East Africa, Bangladesh, India and Nepal. This form tends to affect all age groups.(3)

The pathogenesis of VL involves a complex interaction between the Leishmania parasite, the vector (sandfly) and the immune system of the human host. When the female sandfly injects promastigotes (the mobile, extracellular form) of Leishmania into the skin during a bite, they are rapidly phagocytosed by macrophages and other phagocytic cells. Inside these cells, they transform into amastigotes (intracellular, non-mobile form), capable of reproduction. Infected cells burst, releasing amastigotes that infect other cells. The parasite then migrates to macrophage-rich organs: spleen, liver, bone marrow and lymph nodes.(4)

Although this infection remains asymptomatic and can only be detected by serological tests, in the majority of cases. It is most likely that viable parasites persist after the primary infection, leading to reactivation and disease in cases of immunosuppression such as HIV infection and malnutrition.

The Th1 immune response (interferon-γ, IL-12) is essential for infection control. If this response is inadequate (immunocompromised subjects, HIV), the parasite spreads freely. An excessive or unbalanced immune response can also aggravate the pathology. (3)

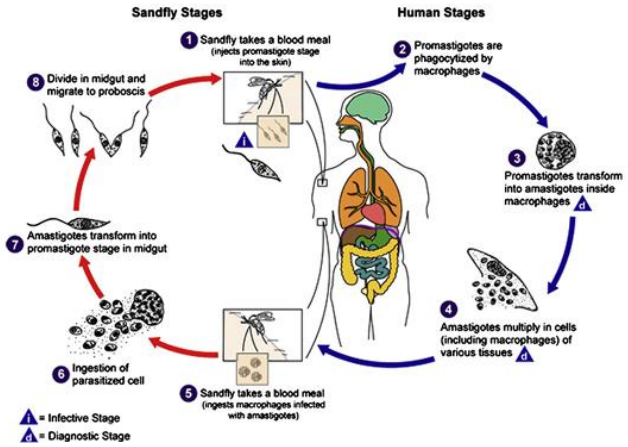

Figure 4 : Life cycle of the leishmania parasite.(3)

VL is often asymptomatic. However, in certain situations, it can be severe, with associated fever, weight loss, hepatosplenomegaly and mood disorders.

Without treatement, VL can result in a high mortality rate. Biological features of VL include cytopenia, hypergammaglobulinemia and the presence of antinuclear antibodies (AN) may resemble the features of certain autoimmune diseases certain autoimmune diseases, in particular systemic lupus erythematosus (SLE).(5)

Oral manifestations of visceral leishmaniasis are rare, but can occur, particularly in immunocompromised patients (especially those with HIV/AIDS) or in reactivated or chronic forms. They are most frequently described in cases of mucocutaneous leishmaniasis, but can also occur in visceral leishmaniasis. The main oral manifestations in VL are deep, painful oral ulcerations, which can resemble aphthous, herpetic or carcinoma lesions. They can be located on the tongue, palate, gums, mucosa jugal and sometimes the pharynx.(6, 7).

Some other manifestations may be noticed, such as gingival inflammation, gingival bleeding secondary to thrombocytopenia, mobility and tooth loss as an indirect consequence of chronic inflammation, secondary infection or osteolysis.

The diagnosis of a first VL infection is established by several ways, including microscopic examination, considered the gold standard allowing the visualization of the amastigote form of the parasite within macrophages by microscopic examination of tissue aspi rates (spleen, bone marrow, or lymphnodes) after Giemsa staining

Other methods are also used, such as a rapid diagnostic test (RDT) detecting antibodies against rK39, the direct agglutination test (DAT) (8)

Three therapeutic options are described in the literature for the treatment of VL : meglumine antimoniate (AM), liposomal amphotericin B (LAMB) and a combination of LAMB and AM. A study was conducted in Brasil in 2019 to assess the cost-effectiveness of treatment strategies for VL. The results analyzed showed that the LAMB strategy is more effective and more cost-effective.(9)

Liposomal amphotericin B is therefore recommended as first-line treatment for VL in Europeand has become the reference treatment in practice according to the World Health Organization (WHO).(10)

Conclusion :

Visceral leishmaniasis, although rare in some regions, remains serious in endemic areas, especially in immunocompromised patients. Oral manifestations, although rare, can be important warning signs. Dentists play a key role in the early detection of these atypical signs. Good clinical knowledge and collaboration with specialists are essential. This enables rapid diagnosis and better patient management.

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