Lip Leishmaniasis with Oral-mucosal Involvement: A Case Report

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Abstract

Statement of the problem: Nowadays leishmaniasis is a common infectious disease around the world which is caused by a protozoan parasite named Leishmania. This parasite is transmitted to human by an infected female sand fly. This disease has three clinical forms; mucosal forms are rare in Iran. **Purpose:** The purpose of this study was to report a case of leishmaniasis with lip and oral mucosal involvement that healed successfully. **Results:** This article is about patients with lip leishmaniasis with oral-mucosal involvement, clinical features were diffuse lip swelling and ulcer in right lower lip vermilion and buccal mucosa. A smear was caught which proved the diagnosis of leishmaniasis. Thereafter, the patients were referred to a dermatologist for treatment and follow up showed complete healing of the lesions. **Conclusion:** Oral mucosal leishmaniasis is rare. However, the swelling may cause different complications like loss of teeth or respiratory obstruction. It should be noted that time is a key point in treatment. So, it is necessary for us to train the differential diagnosis of chronic ulcerated oral lesions to our dentists.

Key words: Lip, Buccal, Leishmaniasis, Swelling, oral-mucosal.

Introduction

Leishmaniasis is a vector borne disease caused by different types of protozoan species of the genus Leishmania, and one of the most important infectious and parasitic diseases in the world.(1) More than 12 million people in 5 continents are affected by leishmaniasis, seen in nearly 100 countries.(2) Because of urbanization, climate change and migration, the number of people who catch this disease is rapidly increasing.(3) Soldiers, travelers who stay in endemic areas, immunosuppressed patients like HIV positive or people who have organ transplant are the most risky group. (1)

Leishmania species are usually separated into Old World and New World types according to their geographic location. Old World Leishmaniasis is found at some parts of Asia, the Middle East, Africa (the continent and countries that have desert) and Southern Europe. New World Leishmaniasis is seen in forested and wet areas, chiefly in some parts of Mexico and Central and South America. Leishmania major, Leishmania tropica and Leishmania aethiopica cause the Old World Leishmaniasis, whereas New World Leishmaniasis are caused by Leishmania mexicana, Leishmania venezuelensis, Leishmania amazonensis or the Viannia complex such as Leishmania brasiliensis, Leishmania panamensis, Leishmania guyanensis and Leishmania peruviana. (3) (Table 1)

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Table 1. Leishmaniasis classification

Clinical form		Species	Geographic area
Cutaneous leishmaniasis (CL)	Old world	L. major	Near East, Africa
		L. tropica	Near East,
		L. aethiopca	Ethiopia, Kenya
		L. infantum	Mediterranean
		L. mexicana complex	Mexico, Central America
		L. braziliensis complex	Brazil, Bolivia
		L. amazonensis	Brazil
	New world	L. braziliensis complex	
Mucocutanesos Leishmaniasis (ML)	Diffuse (anergic) cutaneous Leishmaniasis		
Visceral Leishmaniasis (VL)		L. donovani	India, Kenya
		L. infantum	Mediterranean

The vectors of Leishmania are sandflies that live in different regions like tropics, savannahs and deserts. The size of the insects is between 2 to 3 millimeters, so they can pass through ordinary mosquito nets. (3) The species of sandflies that live in Europe, North Africa, the Middle East and Asia are Phlebotomus and species that spread form southern USA to northern Argentina are Lutzomyia. (4) This insect is active from midnight and its activation depends on whether, in temperate regions the peak of their activity is in the middle of summer. (5)

There are three main clinical forms of leishmaniasis: visceral (VL), cutaneous (CL) and mucocutaneous (ML). (6) Most of the cutaneous forms occur in Middle East countries like Iran, Saudi Arabia and Brazil. Muccosals are frequent in Brazil, Peru and Mexico. (7)

Mucosal involvement in leishmaniasis is rare and the result of haematogenous or lymphatic diffusion of amastigotes from the skin to the oral mucosal. In most cases, ML appears a few years following the emergence ofpreceding cutaneous lesions, but it can develop while the skin lesions are still present. (7)

Oral involvement in Leishmaniasis may have different complications such as loss of teeth or respiratory obstruction that may be fatal. (8) In some cases, voice disorder has been detected. (11) ML can be confused with other diseases such as fungal infections or oral cancer and may be an early or only sign of an immune deficiency disease, for example HIV infection. If detected early, ML can be easily treated before irretrievable damage to oral and nasal tissues. Proper identification helps us to provide good treatment. (8)

Clinical diagnosis of typical wounds is easy, and we can use smear, culture in the test environment NNN (Novy, MacNeal, Nicole) and as the best method, PCR, to diagnose cutaneous leishmaniasis. (9)

Report of Cases

A 12-year-old, generally healthy boy, living in Mashhad, Iran (endemic area), complained of persistent lip enlargement and painful sores present for 6 months, without any fever. The lesion had a granulomatous appearance and fibro-elastic consistency, with slight pain and no bleeding upon palpation, measuring approximately 5.0 · 4.0 cm in its largest diameters. The patient had been receiving treatment for Herpes Simplex with Aciclovir for 5 months. The size of the sore was 2-3 millimeters and became larger after treatment with Aciclovir. (Fig. 1) Oral examination revealed inflammation on the right buccal mucosal that appeared few days after the appearance of lip sores (Fig. 2). We couldn't find any sign of visceral involvement in other organs Such as granulomatous lesions like Sarcoidosis, Crohn, Wegner granulomatous, Cancers, fungal diseases and Leishmania. (18, 19) Due to the high prevalence of Leishmaniasis in the Khorasan Provenance (20), we suspected Leishmaniasis.

For definitive diagnosis, we took skin smears stained with Giemsa.The samples showed Leishman bodies (amastigotes) in macrophage cells, also the smears showed the black pigmentation in macrophage as well as Leishmania bodies.

The patient was referred to Imam Reza Hospital skin section, and was treated with parenteral antimonial sodium stibogluconate, despite the risk of lip deformation; the patient was noncompliant with Additional treatment. Complete healing with no scarring was seen after finishing her treatment process. (Fig. 3)

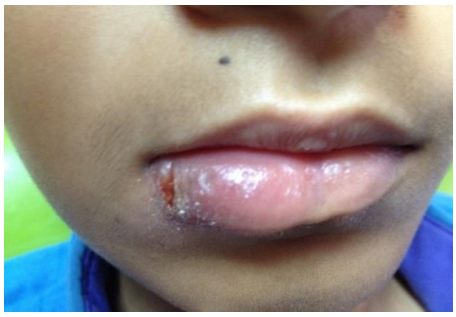

Figure 1. Appearance of the lesion on lip

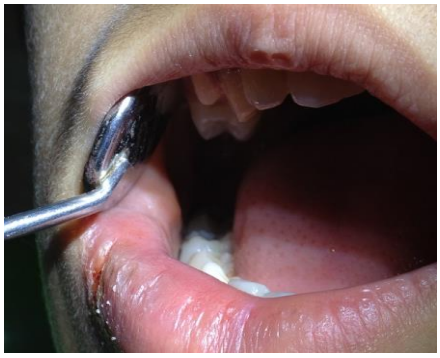

Figure 2. Appearance of the lesion on buccal mucosal

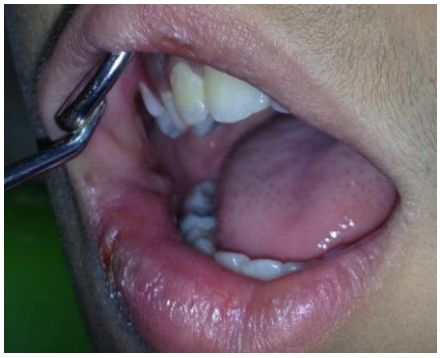

Figure 3. Relative healing of buccal lesion

Discussion

Leishmaniasis is a group of infectious diseases caused by protozoan parasites of the genus Leishmania. This disease has spread around the world and millions of people have been affected by this disease. In recent year,s because of the growing number of patients with immune depression secondary to chronic illness, cancers, transplant, immunosuppressive treatment and HIV infection, the number of Leishmaniasis patients are increasing. (6)

The present article reports another case of oral mucosal leishmaniasis in a young boy whw lived in Mashhad, north-east of Iran. The oral mucosal leishmaniasis is very rare in countries like Iran. (3)

Mucosal leishmaniasis is a disease mainly caused by L. brasiliensis infection, although it has occasionally been registered in infections caused by other Leishmania species like L.tropica. (10) Although the strain of Leishmania is not specified, due to the high prevalence of L.tropica in the Khorasn province and the clinical view of the lesions, it seemed to be L.tropica. (20, 21)

Mucosal involvement in leishmaniasis is rare and the result of haematogenous or lymphatic diffusion of amastigotes from the skin to the oral mucosal. In most cases, ML appears after some years from the emergence of the preceding cutaneous lesions, but it can develop while the skin lesions are still present. (7) One study showed that primary oral leishmaniasis can mimick oral cancer. (17) In this case, we understood that the oral swelling appeared a few days after the appearance of lip sores.

The clinical aspect of the mucosal lesion was similar to that reported for other infectious or non-infectious diseases, such as herpes simplex, Actinomycosis, Blastomycosis, Histoplasmosis, leprosy, granulomatous lesions and squamous cell carcinoma or allergic responses, which have different treatments and prognoses.

In the history of the patient, we couldn't find an evidence of an allergic response. Patient's oral hygiene was at a good level. On the other hand, the treatment for herpes simplex wasn't any effective on sores. Fungal infections are rare too and are usually manifested as painful gingival ulcers, gray-colored diffusely swollen peripheral mucosa and erythema nodosum (21, 22). As a result, granulomatous lesions were our first recognition and between all granulomatous lesions, the most likely were Crohn's disease, Sarcoidosis, Wegener's granulomatosis, deep fungal infections and Leishmaniasis. (18) Leishmaniasis is the best diagnosis at the first look, because of the clinical appearance of the lesion, patient home address and existing leishmaniasis in other family members.

Crohn's disease has Aphthous-like lesions, mucosal overgrowth with cobblestone appearance, small mucosal postules and deep linear ulcers. Sarcoidosis shows solitary or multiple gingival nodules, xerostomia, osseous involvement, salivary glands, facial nerve palsy. Finally, Wegener's granulomatosis has Strawberry

gingivitis, palatal ulcer and facial nerve palsy. (18, 19, 23)

Due to the presence of lip lesions and oral inflammation we suspected mucosal leishmaniasis and reloaded others.

Some cases show that this form of leishmaniasis appears months or even years after the spontaneous or therapeutic cure of the primary cutaneous lesions and is associated with delayed healing and failure to treat primary cutaneous lesions (13). Spontaneous reactivation (14, 15) and transmission along with organ transplants (16) is the most important reason for persistence of Leishmania parasites in human body. In this article, the patient reported no previous cutaneous lesions or immunosuppression. As a result, we diagnosed mucosal leishmaniasis.

For the first diagnostic action, the skin smear with Giemsa was done, and leishmaniasis was confirmed and the patient was referred to Imam Reza Hospital skin section for treating.

There are many different treatment methods for leishmaniasis such as injection of Meglumine antimoniate at 20 mg/kg per day dose until complete healing, Cryotherapy, thermotherapy and removing the lesion with surgical techniques. (12)

The method of choice in treatment of mucosal leishmaniasis is Meglumine antimoniate with a dose of 20 mg/kg per day in 50 mL of 5% dextrose for 4 weeks.

Conclusion

In conclusion, oral mucosal leishmaniasis is a rare lesion. However, the lesion may cause different complications such as loss of teeth or respiratory obstruction. It should be noted that time is a key point in success of treatment. So it is necessary for us to train all differential diagnoses of chronic ulcerated oral lesions to our dentists.

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