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Lip leishmaniasis: evaluation of 20 patients

Dudakta leishmaniasis: 20 hastanın değerlendirilmesi

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Abstract

Objective Mucosal leishmaniasis (ML) is an important public health problem because it has a significant morbidity and mortality rate in undeveloped countries. In this study, clinical features of patients diagnosed with ML in Sanliurfa, an endemic region for leishmaniasis, were evaluated.

Methods In this retrospective study, patients admitted to the skin and venereal diseases clinics of two different training and research hospitals between May 2015 and September 2019 and diagnosed as ML by microscopic examination were included.

Results In this study, 446 patients with CL were retrospectively evaluated and 24 lesions of 20 patients with lip involvement were included. Of the 20 patients included in the study, 11 (55%) were male and 9 (45%) were female. Lesions were seen only in the lips in 15 (75%) patients, while additional skin involvement was present in 5 (25%) patients. None of the patients had gingival or genital involvement.

Conclusion In conclusion, ML should be considered when treatment resistant lesions develop in the labial region of the patients living in endemic areas or travelling to endemic areas and the diagnosis should be confirmed and treated early.

Key words: *lip, cutaneous leishmaniasis, mucosal leishmaniasis*

Özet

Amaç Mukozal leishmaniasis (ML) gelişmemiş ülkelerde önemli bir morbidite ve mortalite oranına sahip olduğu için önemli bir halk sağlığı sorunudur. Bu çalışmada leishmaniasis için endemik bir bölge olan Şanlıurfa ilinde ML tanısı konulan hastaların klinik özellikleri değerlendirildi.

Yöntem Bu retrospektif çalışmamıza, iki ayrı eğitim ve araştırma hastanesinin deri ve zührevi hastalıkları kliniğine Mayıs 2015- Eylül 2019 tarihleri arasında başvuran ve mikroskobik incelemeyle ML tanısı konulan hastalar dahil edildi.

Bulgular Çalışmamızda 446 KL hastası retrospektif olarak incelendi, dudak mukozası tutulumu olan 20 hastanın 24 lezyonu dahil edildi. Çalışmaya katılan 20 hastanın 11'i (%55) erkek, 9'u (%45) kadındı. 15(%75) hastada

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lezyonlar sadece dudaklarda görülürken, 5(%25) hastada ayrıca deri tutulumu da mevcuttu. Hiçbir hastada diş eti tutulumu ve genital bölge tutulumu yoktu.
Sonuç Sonuç olarak leishmaniasis için endemik olan bölgelerde yaşayan veya bu bölgelere seyahat eden kişilerde dudak bölgesinde tedaviye dirençli lezyonlar geliştiğinde ML düşünülmeli ve hastalığın tanısı doğrulanıp erken dönemde tedavi edilmelidir.

Anahtar kelimeler: *dudak, kutanöz leishmaniasis, mukozal leishmaniasis*

Introduction

Leishmaniasis is a complex of diseases caused by the bite of a female sand fly vector which is infected with the microorganisms of leishmania protozoan. This infection occurs as cutaneous leishmaniasis (CL), mucocutaneous leishmaniasis (MCL) or visceral leishmaniasis (VL) depending on the type of leishmania and host immune response.[1-5] CL is grouped into old-world and new-world CL according to geographic region.[6,7] Mucosal leishmaniasis (ML) is endemic in South America and is caused by L. brasiliensis.[8-10] ML is an important endemic disease and public health problem as it has a significant morbidity and mortality rate in undeveloped countries.[11]

In this study, clinical features of patients diagnosed with ML in Sanliurfa, an endemic region for leishmaniasis, were evaluated.

Methods

In In this retrospective study, patients admitted to the skin and venereal diseases clinics of two different training and research hospitals between May 2015 and September 2019 and diagnosed as ML with lip involvement by microscopic examination were included.

Clinical and sociodemographic data were recorded such as age, sex, number of lesions, location of lesions, size and duration of lesions, presence of skin involvement other than mucosal involvement, survival in an endemic region, intralesional or systemic antimony therapy.

Statistical analysis were performed using SPSS 21.0 (SPSS Inc., Chicago, IL, USA). Continuous data were calculated as mean ± standard deviation (SD) and categorical data were calculated as frequency (%). Ethics committee approval was received from our hospital for the study.

Results

In this study, 446 CL patients were evaluated retrospectively and 24 lesions of 20 patients with lip involvement were included. Of the 20 patients included in the study, 11 (55%) were male and 9 (45%) were female. The mean age of the patients was 22.80 ±6.27 years. 15 (75%) patients had a history of survival in the endemic region. Six (25%) lesions were papules and 18 (75%) lesions were plaque (Fig. 1). Lesions were seen only on the lips in 15 (75%) patients while

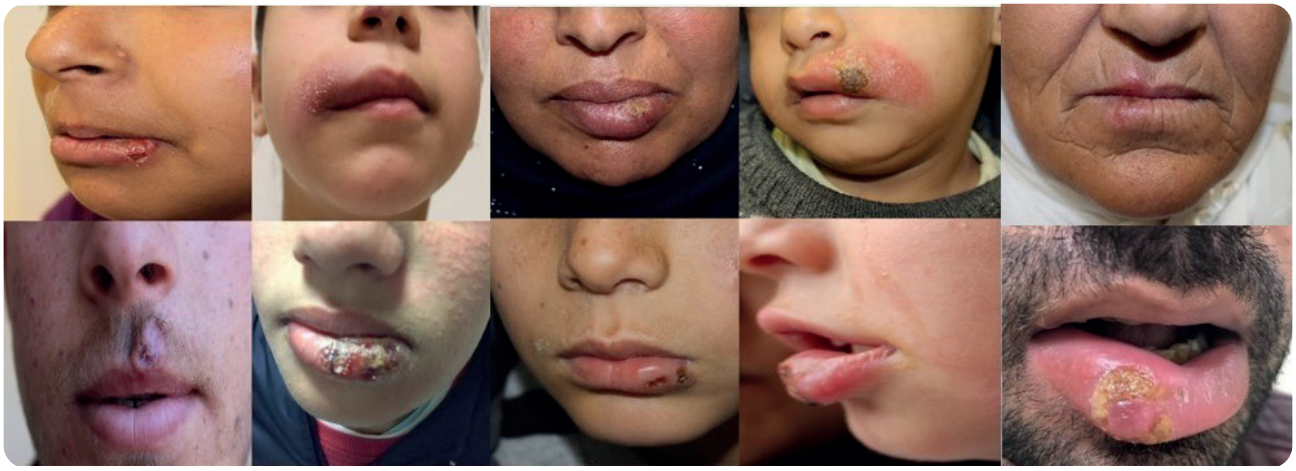

Fig. 1. Leishmania lesions located on the lips

skin involvement concurrent with lip involvement was present in 5 (25%) patients. None of the patients had gingival or genital involvement. The mean lesion duration was 4.30 ± 3.70 months. The mean lesion size was 2.30 ± 3.10 cm. All patients underwent cutaneous smears and were diagnosed by microscopic examination. (Fig. 2) After the diagnosis, 12 patients (60%) received sysemic antimony and 8 patients (40%) received intralesional antimony. In 15 patients, it was seen that the lesions healed with minimal scarring and the treatment response of 5 patients could not be evaluated because they did not come to follow up.

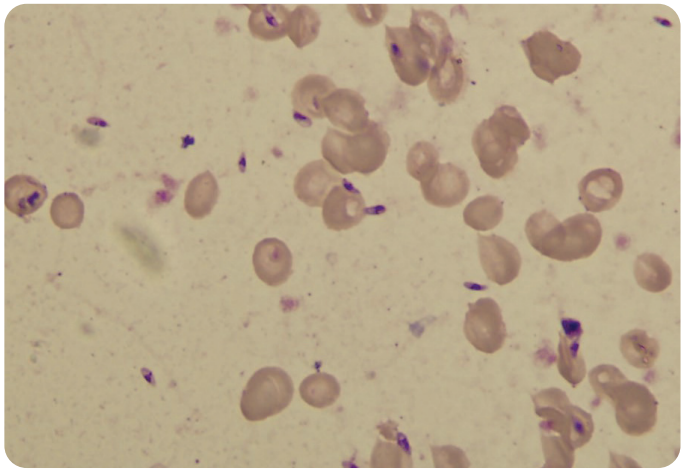

Fig. 2. Leishmania amastigotes seen from samples taken from a lesion (Giemsa, 100X)

Discussion

ML is frequently seen in new world leishmaniasis, whereas mucosal involvement in old world leishmaniasis is limited.[8-10] Kharfi et al. reported 5 ML patients with lip involvement in four and endonasal mucosa involvement in one patient.[10] El-Hoshy et al. reported 12 ML cases with localized lesions on the lips.[12] Sitheeque et al. reported 492 patients with ML localized to the perioral regions and lips in 1990.[13] In a retrospective study of 14400 CL patients in Sanliurfa province by Yesilova et al. lip involvement was found to be 4.3% (621 patients). In addition, 71.9% (447 patients) of the patients with mucosal involvement had concomitant cutaneous involvement. None of the patients had visceral involvement. In only one of the patients, gingival involvement was also detected. In our study, lesions were only seen on the lips in 15 (75%) patients, while skin involvement concurrent with lip involvement was present in 5 (25%) patients. None of the patients had gingival and genital involvement.

Lip leishmaniasis is characterized clinically by the gradual and proceeding expansion of one or both lips and macrocheilitis is the final presentation. A papule, nodule or plaque often demonstrate within the swelling undergoes an ulceration which may be covered by crusts and scaling. The consistency of the entire lesion is parenchymatous-hard.[15-18] In the study of Yesilova et al., the mean lesion diameter and number were 14.37 ± 11.83 mm and 1.91 ± 1.45, respectively, and ulcerative lesions were the most common (68.44%).[14] In our study, the mean lesion size was 2.30 ± 3.10 cm and the most common lesion type was plaque (75%).

Lip involvement may result from direct spreading from nearby skin lesions or the spread of Leishmania amastigotes through the hematogenous or lymphatic pathway.[19] Patients with lip involvement may be of any age and have good overall health, but cervical lymphadenopathy may also occur.[20] In the study of Yesilova et al., lip involvement was more common in female patients and the mean age of the patients was found to be 15.34 ± 15.26.[14] In our study, lip involvement was more common in male patients and the mean age of the patients was 12.80 ± 6.27 years.

Lip lesions may mimic herpes labialis, Crohn's disease, sarcoidosis, Melkersson-Rosenthal syndrome, basal cell carcinoma, squamous cell carcinoma, and mycotic infections.[21-25] For the diagnosis of ML, parasitological confirmation is required in patients with a history of living in the endemic areas or travelling to the endemic areas and having clinical findings suggestive of ML.[26] The diagnosis is confirmed by the presence of amastigotes on microscopic examination of smears stained with Giemsa. Amastigotes are abundant in early lesions, whereas amastigotes are rare in late and secondary infected lesions. Amastigotes are difficult to detect in chronic cases due to their scarcity.[1,27] In addition;

fine needle aspiration method, culture, incisional skin biopsy and polymerase chain reaction (PCR) method with biopsy material or samples sent from skin aspirates are used.[26] In the study of Yesilova et al. the diagnosis of ML was made by cutaneous smear, culture and histopathological examination.[14] In our study, the diagnosis of ML was made by cutaneous smear in all patients. Lip lesions can be treated with intralesional antimony, systemic antimony or combinations depending on the clinical symptoms. Intralesional antimony therapy may cause a burning sensation, pain, and vasovagal reactions.[1,20] Other therapeutic agents used in the treatment of ML include liposomal amphotericin B, pentamidine, oral azole compounds and miltefosine.[28-30] In a ML case reported in Sudan, upper lip, larynx, palate and gingiva involvement was detected and this patient was successfully treated with intravenous sodium stibogluconate.[9] Kharfi et al. successfully treated 5 ML patients with intramuscular meglumine antimoniate therapy.[10] In the study of Yesilova et al. all patients were treated with intralesional antimony.[14] In our study, systemic antimony was given to 12 (60%) patients and intralesional antimony treatment was given to 8 (40%) patients.

The limitations of our study were the inability to detect leishmania species due to lack of PCR and the lack of treatment response in 5 patients.

In conclusion, ML disease should be considered when treatment resistant lesions develop in the lip region in people living in endemic areas or traveling to endemic areas and the diagnosis should be confirmed and treated early. Prospective studies with a large number of patients are needed to better understand the clinical manifestations of ML.

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