



CLINICAL STUDY

AURICULAR CUTANEOUS LEISHMANIASIS: A RETROSPECTIVE EVALUATION OF 12 PATIENTS

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SUMMARY

Aim: Leishmaniasis refers to a group of diseases caused by protozoan parasites belonging to the genus *Leishmania*. Cutaneous leishmaniasis (CL) lesions are typically observed as single or multiple lesions, particularly in the head-neck region. Auricular involvement in CL is extremely rare. The aim of this study is to examine the clinical features, diagnosis, and treatment methods of 12 patients diagnosed with auricular CL.

Material and methods: In this retrospective study, the files of 12 patients diagnosed with CL through clinical and microscopic examination, and presenting with auricular involvement, were reviewed at our Dermatology and Venereology clinic between October 2019 and October 2022. Clinical and demographic features such as age, gender, number, localization, size, and duration of lesion, and treatments received were recorded from the patients' files.

Results: All patients diagnosed with CL were male (100%). The mean age of patients was 15.41 ± 14.94 . All patients presented with involvement of a single ear. Helix involvement was observed in 6 (50%) patients, antihelix in 4 (33%) patients, and lobule in 2 (17%) patients. The average duration of lesions was 4.6 ± 2.8 months. The average size of the lesions was 3.4 ± 2.3 cm. Cutaneous leishmaniasis diagnosis was made by direct microscopic examination in all patients, and all were treated with meglumine antimoniate (MA) at a dose of 20 mg/kg/day for 20 days intramuscularly. No cutaneous or systemic side effects related to MA treatment were observed in any patients.

Conclusion: The possibility of auricular involvement in CL should be kept in mind, and the diagnosis should be confirmed by microscopic examination. Systemic MA treatment should be considered in patients with auricular region involvement.

Keywords: Auricle, Cutaneous Leishmaniasis, meglumine antimoniate, smear

AURİKULER KUTANÖZ LAYŞMANYAZ: 12 HASTANIN RETROSPEKTİF DEĞERLENDİRİLMESİ ÖZET

Amaç: Layşmanyaz, *Leishmania* genusuna ait protozoan parazitlerin neden olduğu bir dizi hastalığı tanımlamaktadır. Kutanöz layşmanyaz (KL) lezyonları özellikle baş-boyun bölgesinde tek veya çoklu lezyonlar şeklinde görülmektedir. Kutanöz layşmanyaz" de aurikuler bölge tutulumu son derece nadirdir. Bu çalışmadaki amacımız aurikuler KL tanısı konulan 12 hastanın klinik özelliklerini, tanı ve tedavi yöntemlerini incelemektir.

Yöntem ve Gereçler: Bu retrospektif çalışmada Deri ve zührevi hastalıkları kliniğimizde Ekim 2019 ? Ekim 2022 tarihleri arasında klinik ve mikroskopik inceleme ile KL tanısı konulan ve aurikula tutulumu olan 12 hastanın dosyaları retrospektif olarak incelendi. Hastaların dosyalarında kayıtlı olan yaş, cinsiyet, lezyon sayısı, lezyon lokalizasyonu, lezyon büyüklüğü, lezyonun süresi ve aldıkları tedaviler gibi klinik ve demografik özellikler kaydedildi.

Bulgular: Kutanöz layşmanyaz tanılı hastaların tamamı (%100) erkek idi. Hastaların yaş ortalaması 15.41 ± 14.94 idi. Bütün hastalarda tek kulak tutulumu mevcuttu. 6 (%50) hastada heliks tutulumu, 4(%33) hastada antiheliks tutulumu ve 2(%17) hastada lobül tutulumu mevcuttu. Ortalama lezyon süresi 4.6 ± 2.8 ay idi. Ortalama lezyon büyüklüğü 3.4 ± 2.3 cm idi. Kutanöz layşmanyaz tanısı tüm hastalarda direk mikroskopik inceleme ile konuldu ve tüm hastalara meglumin antimonate (MA) 20 mg/kg /gün dozunda 20 gün boyunca IM olarak uygulandı. Hiçbir hastada MA tedavisine bağlı kutanöz ve sistemik yan etki görülmedi.

Sonuç: Kutanöz layşmanyaz" de aurikuler bölge tutulumu olabileceği akılda tutulmalı ve mikroskopik inceleme yapılarak hastalığın tanısı doğrulanmalıdır. Aurikuler bölge tutulumu olan hastalarda sistemik MA tedavisi düşünülmelidir.

Anahtar Sözcükler: Aurikula, kutanöz layşmanyaz, meglumin antimonate, smear

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INTRODUCTION

Cutaneous leishmaniasis (CL), caused by *Leishmania* protozoa, is a disease transmitted through the bites of infected *Phlebotomus* genus sandflies¹. Although CL has a wide variety of clinical presentations, the most common form is ulcerative crusted nodules and plaques that heal

spontaneously leaving scars²⁻⁴. Treatment in CL varies according to the location of the lesion, its size, and the patient's immune status. Treatment goals are to minimize local damage and functional losses, accelerate healing, and reduce the possibility of recurrence. The most preferred drugs in the treatment of CL are pentavalent antimony compounds such as meglumine antimonate and sodium stibogluconate^{1,2,5}. Lesions in CL are most frequently seen in the head-neck region as single or multiple lesions⁵⁻⁷. A type of ear leishmaniasis caused by *Leishmania mexicana*, commonly found in Mexico and Central America, is known as "Chiclero's ulcer," primarily affecting the ears of

forest workers. However, ear leishmaniasis in the Old World is quite rare^{8,9}. Cutaneous leishmaniasis in this location can be mistaken for malignancies, other infectious diseases, and inflammatory processes. Therefore, particularly in endemic areas for CL, cutaneous leishmaniasis should be considered in the differential diagnosis of ulcerative lesions and auricular enlargements⁸⁻¹¹. Despite its rarity, delayed diagnosis and treatment of auricular involvement can lead to destruction in the ear^{9,12-14}.

The aim of this study is to examine the clinical characteristics, diagnosis, and treatment methods of 12 patients diagnosed with auricular CL between October 2019 and October 2022.

MATERIAL and METHODS

In this retrospective study, the files of patients diagnosed with CL through clinical and microscopic examination, were reviewed at our Dermatology and Venereology clinic between October 2019 and October 2022. The files of 1784 patients diagnosed with CL were examined retrospectively, and 12 patients with auricular involvement and no missing data in their files were included in the study.

Clinical and demographic features such as age, gender, number, localization, size, and duration of lesion, and treatments received were recorded from the patients' files. Lesion size was determined by measuring the diameter of the lesion. Diagnosis of CL was made by observing amastigotes in dermal scrapings from solid lesions like papules, nodules, plaques, and in smear materials from crusted ulcerative lesions. Systemic meglumine antimonate (MA) therapy was administered at a dose of 20 mg/kg/day for 20 days intramuscularly (IM). In patients treated with systemic MA, hemogram, lipase, amylase,

and renal and liver function tests were checked weekly for the first week and then bi-weekly. Additionally, electrocardiograms were conducted every other day in the first week and then twice a week.

Statistical analyses were performed using SPSS 21.0 (SPSS Inc., Chicago, IL, USA). Continuous data were expressed as mean \pm standard deviation (SD), and categorical data as frequency (%).

Ethical approval for the present study was obtained from the local ethics committee (Number: 23.20.23/2023).

RESULTS

All patients diagnosed with CL were male (100%). The mean age of patients was 15.41 ± 14.94 . All patients presented with involvement of a single ear. Helix involvement was observed in 6 (50%) patients, antihelix in 4 (33%) patients, and lobule in 2 (17%) patients (Figure 1). The average duration of lesions was 4.6 ± 2.8 months. The average size of the lesions was 3.4 ± 2.3 cm. CL diagnosis was confirmed by direct microscopic examination in all patients. (Figure 2) All patients were treated with IM MA at a dose of 20 mg/kg/day for 20 days. No cutaneous or systemic side effects related to MA treatment were observed in any patients. Complete healing of lesions was observed in all patients after 12 weeks of treatment.



Figure 1: A) Crusted and erythematous plaque lesion, B) Crusted and erythematous plaque lesion, C) Crusted papular lesion, D) Erythematous plaque lesion, E) Erythematous telangiectatic patch, F) Keloidal lesion.

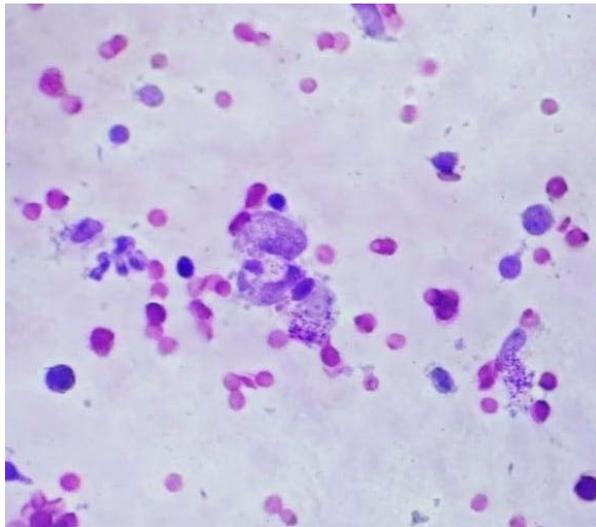


Figure 2: Amastigotes are seen inside and outside of macrophages cells of CL lesion smears stained with Giemsa (Giemsa x40)

DISCUSSION

In Türkiye, a country endemic for CL, the province of Şanlıurfa is a hyperendemic region with the highest number of reported cases^{3,6}. Cutaneous leishmaniasis can be seen in all age groups, but it is most commonly observed in children and adolescents³⁻⁶. In the present study, 58% (7/12) of the patients were under 18 years old.

Cutaneous leishmaniasis is reported at a higher rate in women in endemic regions. Previous studies in our country have also shown a female predominance¹. Aksoy et al. reported that of 8786 pediatric patients, 4050 (46.1%) were male and 4736 (53.9%) were female⁶. However, studies in other endemic countries like Iran and Tunisia with pediatric patients have reported a male predominance^{15,16}. In the present study, all patients (100%) were male. This may be related to the practice of young girls wearing headscarves from an early age in the region and the freedom of boys to be outdoors during the hours when most bites occur.

In the present study, the average duration of lesions was 4.6 ± 2.8 months. Aksoy et al. reported the average duration of lesions as 8.58 weeks⁶. The longer duration of illness in the present study could be related to the rarity of auricular involvement in CL, leading to a

delayed diagnosis. The average lesion size in the present study was 3.4 ± 2.3 cm, while Aksoy et al. reported an average lesion diameter of 12.77 ± 0.11 mm⁶. The larger size of lesions in the present study could be due to the delayed diagnosis, allowing lesions to grow larger over time.

Cutaneous leishmaniasis lesions usually arise from bites of infected female sandflies, particularly in body areas not covered by clothing^{6,12}. The head and neck region is the most affected area. In Türkiye, studies have reported head and neck involvement rates of 60.7% and 57.3%, respectively^{12,13}. Similar results were observed in studies focusing solely on pediatric populations in our country and Iran^{5,6,14,15}. When the head and neck region is examined by compartments, the cheek is the most commonly affected site^{5,13,14,16,17}. Studies have reported either no ear involvement or ear involvement rates of 6.6%, 4.6%, and 1%^{5,13,17}. The auricle anatomically consists of three parts: helix, antihelix, and lobule, and case reports in the literature have described involvement of each part separately¹⁸⁻²¹. In the present cases, helix involvement was found in 6 (50%) patients, antihelix in 4 (33%) patients, and lobule in 2 (17%) patients. The high frequency of helix involvement could be related to its position and surface area making it more susceptible to bites.

While ear leishmaniasis is rare in the Old World, the New World has a type known as "chiclero ulcer," most commonly affecting the pinna. It is typically seen among forest workers (chicleros) in Mexico and Central America, where *Leishmania mexicana* is usually the causative agent⁸. An epidemiological study in Mexico found the annual incidence of the disease to be 508 cases per 100,000 individuals¹⁸.

The first step in diagnosing CL is clinical suspicion. While typical lesions in endemic areas easily bring CL to mind, in non-endemic areas, a high index of suspicion is required^{1,2}. Laboratory methods are then needed to confirm the diagnosis. Due to its high specificity, parasitological diagnosis is still considered the gold standard. Cutaneous smear, culture, incisional biopsy, and PCR are methods aimed at



demonstrating the parasite. Among these, cutaneous smear is a valuable method, especially in endemic areas, because it is cheap, quick, and easy to apply²²⁻²⁵. In the present patients, we confirmed the clinical diagnosis by observing amastigotes using the cutaneous smear method.

Cutaneous leishmaniasis lesions spontaneously heal, leaving an atrophic scar at the site. However, treatment is recommended to hasten healing, prevent unsightly scarring, prevent functional impairment, avoid relapses, prevent mucosal involvement via local spread, and remove the patient from being a disease reservoir. In treatment, antifungal agents like pentavalent antimony compounds, amphotericin-B, ketoconazole, fluconazole, itraconazole, terbinafine, pentamidine, miltefosine, paromomycin, and allopurinol, as well as physical methods like cryotherapy, thermotherapy, and photodynamic therapy can be used^{1,2,25,26}. Pentavalent antimony compounds (MA, sodium stibogluconate) continue to be the first treatment choice in most countries. However, the choice of appropriate treatment often depends on the clinician's experience, patient preferences, and cost-effectiveness considerations for the patient and/or the health system²⁵. We preferred pentavalent antimony compounds in all of the present cases, patients, not only to prevent the development of ugly cicatrix, destructions and dysfunction by getting a rapid response, but also because they can be obtained free of charge from the Oriental Furuncle Diagnosis and Treatment Center in our province.

These drugs can be administered both intraleisonally and systemically. Systemic therapy is recommended in chronic, recurrent, or relapsing lesions, lesions over 5 cm, inflammatory lesions, more than five lesions, lesions in locations that can cause functional impairment (over joints, eyelids, lips, etc.), mucosal involvement, proximity to mucosa and cartilage, nodular lymphangitis, immunocompromised states, and in cases unresponsive to local treatments²⁶.

It was choose systemic therapy for all the present cases, patients due to the risk of cartilage

involvement and functional impairment in ear lesions.

According to WHO recommendations, systemic pentavalent antimony treatment for 20 days at a dose of 10-20 mg/kg/day is usually sufficient²⁵. In a Tunisian pediatric series, systemic antimony treatment for 7-15 days at a dose of 20 mg/kg/day was successfully used in pediatric patients with more than five lesions or those with lesions close to the cartilage who did not respond to IL antimony treatment¹⁶. Intralesional treatments can also be used in patients where systemic antileishmanial treatments cannot be used due to contraindications^{2,7} Yaghoobi et al successfully treated a patient with auricular leishmaniasis with intralesional meglumine antimoniate (27. Diociaiuti et al. successfully treated a pediatric patient with auricular leishmaniasis with intralesional liposomal amphotericin B²⁸. We administered systemic MA treatment at a dose of 20 mg/kg/day for 20 days to all the present patients and achieved clinical improvement.

CONCLUSION

In CL, auricular involvement should be kept in mind, and the diagnosis should be confirmed by microscopic examination. Systemic MA treatment should be considered in patients with auricular involvement.

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