CLINICAL STUDY

NECK CUTANEOUS LEISHMANIASIS

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SUMMARY

Aim: Lesions in cutaneous leishmaniasis (CL) commonly occur in uncovered areas of the body, such as the head and neck. CL, one of the major mimicking diseases, should be considered in the differential diagnosis of neck lesions, and laboratory methods should be used to confirm the diagnosis in suspected cases. The aim of this study is to examine the clinical features, diagnostic methods, and treatment options of CL localized to the neck. To the best of our knowledge, there has been no prior study in the literature specifically evaluating neck involvement in CL.

Material and Methods: This retrospective study included 81 patients with CL lesions exclusively on the neck, diagnosed between September 2018 and October 2024 at the Diagnosis and Treatment Center for Oriental Boil in Şanlıurfa, an endemic region for CL. Clinical and demographic characteristics of the patients were retrieved from their medical records.

Results: Of the patients, 74% were male, and 26% were female. The mean age was 20.59 ± 15.86 years, and the majority (63%) were in the pediatric age group. Acute CL was identified in 66 patients, while 15 patients had chronic CL. The mean lesion duration was 11.56 ± 0.79 months. Solitary lesions were present in 63% of the patients, whereas 37% had multiple lesions, with a mean lesion count of 1.8. The mean lesion diameter was 14.38 ± 3.05 mm. The lesion types, in order of frequency, were nodulo-ulcerative, nodular, and papular. A total of 98.8% of patients responded to intralesional meglumine antimonate (ILMA) treatment, while the remaining one patient required systemic meglumine antimonate (MA) therapy.

Conclusion: Clinicians in endemic regions should always consider CL in the differential diagnosis of various types of neck lesions. ILMA is a highly effective treatment for neck CL.

Keywords: Cutaneous leishmaniasis, intralesional meglumine antimonate, neck

BOYUN KUTANÖZ LEİSHMANİASİSİ

ÖZET

Amaç: Kutanöz leishmaniasis (KL) lezyonları genellikle baş ve boyun gibi vücudun açık bölgelerinde görülür. Taklitçi hastalıklardan biri olan KL, boyun lezyonlarının ayırıcı tanısında düşünülmeli ve şüpheli vakalarda tanıyı doğrulamak için laboratuvar yöntemleri kullanılmalıdır. Bu çalışmanın amacı, boyunda lokalize KL'nin klinik özelliklerini, tanı yöntemlerini ve tedavi seçeneklerini incelemektir. Bildiğimiz kadarıyla, literatürde KL'de boyun tutulumunu özel olarak değerlendiren daha önce yapılmış bir çalışma bulunmamaktadır.

Gereç ve Yöntemler: Bu retrospektif çalışmaya, KL için endemik bir bölge olan Şanlıurfa'daki Doğu Çıbanı Tanı ve Tedavi Merkezi'nde Eylül 2018 ile Ekim 2024 arasında teşhis edilen, yalnızca boyunda KL lezyonları olan 81 hasta dahil edildi. Hastaların klinik ve demografik özellikleri tıbbi kayıtlarından elde edildi.

Bulgular: Hastaların %74'ü erkek, %26'sı kadındı. Ortalama yaş 20,59 ± 15,86 yıldı ve çoğunluğu (%63) pediatrik yaş grubundaydı. 66 hastada akut KL saptanırken, 15 hastada kronik KL vardı. Ortalama lezyon süresi 11,56 ± 0,79 aydı. Hastaların %63'ünde soliter lezyonlar mevcuttu, %37'sinde ise çoklu lezyonlar vardı ve ortalama lezyon sayısı 1,8 idi. Ortalama lezyon çapı 14,38 ± 3,05 mm idi. Lezyon tipleri, sıklık sırasına göre nodülo-ülseratif, nodüler ve papülerdi. Hastaların %98,8'i intralezyonel meglumin antimonat (ILMA) tedavisine yanıt verirken, kalan bir hasta sistemik meglumin antimonat (MA) tedavisine ihtiyaç duydu.

Sonuç: Endemik bölgelerdeki klinisyenler çeşitli boyun lezyonu tiplerinin ayırıcı tanısında her zaman KL'yi göz önünde bulundurmalıdır. ILMA, boyun CL için oldukça etkili bir tedavidir.

Anahtar Sözcükler: Kutanöz leishmaniasis, intralezyonel meglumin antimonat, boyun

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Received: 17 December 2024, revised for: 20 February 2025, accepted for publication: 09 May 2025

Cite this article: An İ., Yentur Doni N., Kaya K., Ertürk T., Ceylan M. R., Solmaz A.. Neck Cutaneous Leishmaniasis KBB-Forum 2025;24(2):143-149

INTRODUCTION

Cutaneous leishmaniasis is a vector-borne parasitic disease transmitted via Phlebotomus bites and is endemic in over 90 countries. Türkiye is among the countries where CL is endemic, and Şanlıurfa province reports the highest number of cases. Cutaneous leishmaniasis frequently affects exposed areas of the body; therefore, the head and neck region, along with the extremities, are highly susceptible to lesions[1,2]. Affected areas often exhibit solitary

or multiple lesions, commonly presenting as ulcers, leading to significant consequences such as disfiguring scars, severe disability, and social stigma[3,4].

Clinical suspicion of CL should be confirmed with laboratory methods, with microscopic identification of the parasite being the most commonly used diagnostic approach[5].

Patients should receive treatment after diagnosis to prevent their role as reservoirs for the disease. Pentavalent antimonial compounds are the most commonly used drugs for treatment[6].

The aim of this study is to evaluate the sociodemographic and clinical characteristics, diagnostic methods, and treatment options for CL localized to the neck.

MATERIAL and METHODS

This retrospective study included 81 patients diagnosed with CL lesions exclusively on the neck between September 2018 and October 2024 at the Diagnosis and Treatment Center for Oriental Boil in Şanlıurfa. CL diagnosis was confirmed through direct microscopic examination and histopathological analysis of the affected skin. Positive microscopic findings were defined as smears containing amastigotes with round or oval shapes, a dark purple-stained nucleus at one end, and a kinetoplast adjacent to it, with a pale blue cytoplasm. Positive histopathological findings were defined as preparations stained with hematoxylin and eosin or Giemsa showing amastigotes. Patient data, including gender, age, disease duration, lesion count, lesion diameter, lesion types, microscopic examination results, treatments received, and treatment responses, were collected from medical records.

Patients diagnosed with CL received intramuscular meglumine antimonate (IMMA) or intralesional meglumine antimonate (ILMA). IMMA was administered at a dose of 20 mg/kg/day for 21 days, while ILMA was administered twice weekly for 4 weeks.

Inclusion and Exclusion Criteria

Inclusion criteria included being aged 0-80 years, having a diagnosis of cutaneous leishmaniasis (CL) confirmed by microscopic or histopathological examination, and having neck involvement.

Exclusion criteria were incomplete medical records, lack of diagnostic confirmation through microscopic or histopathological examination, absence of neck involvement, and having lesions in areas other than or in addition to the neck.

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008. Ethics committee approval was granted from Harran University Medical Faculty Hospital on 29.004.2024 with protocol number HRU.24.05.33.

Statistical Analysis

Patient data collected within the scope of the study were analysed with the IBM Statistical Package for the Social Sciences (SPSS) for Windows 20.0 (IBM Corp., Armonk, NY) package program. Frequency and percentage for categorical data and mean and standard deviation for continuous data were given as descriptive values.

RESULTS

The mean age of all individuals was 20.59 ± 15.86 years (range = 4-77 years), and 74.13% were male. Of the patients, 63% (n=51) were under 18 years of age. Sixteen percent were aged 19-36 years, 19.8% were aged 37-54 years, and only one patient was over 54 years old.

A diagnosis of CL was established by direct microscopic examination of affected skin in 75 patients (92.5%) and by histopathological examination in 6 patients (7.5%).

The year with the highest diagnosis rate of neck CL cases was 2021 (34.6%), while the lowest rates were in 2023 and 2024 (3.7%). Throughout all years, male patients predominated (Figure 1).

When all lesions were assessed according to their clinical appearances, the most common lesion type was nodulo-ulcerative (61.7%), followed by nodular (28.0%) and papular lesions (10.3%) (Table 1).

When lesion counts were analyzed by age groups, solitary lesions were the most frequent in all age groups. Of the all CL patients 63,0% (51/81) had one lesion, 19,8 % (16/81) had two lesions, 7,4% (6/81) had three lesions, 4,9% (4/81) had four, 1,2% (1/81) had 5 lesions, 1,2%

(1/81) had 6 lesions and 2,5% (2/81) had 9 lesions (Table 2).

The mean number of lesions was 1,8 ± 1,5 (range 1-9). The average lesion size (longest axis) was 14,38 ± 3,05 mm (range 5-60 mm) and the average lesion duration (month) was 11,56±0,79 (range 4-40) (Table 3).

Acute CL was present in 81.5% of the patients, while 18.5% (n=15) had chronic disease.

Therapy of CL cases

%98,8 (n=80) of neck CL patients recovered after receiving only one course of IL MA (8 total injections of IL). %1,2 (n=1) of the CL patients were administered systemic MA therapy.

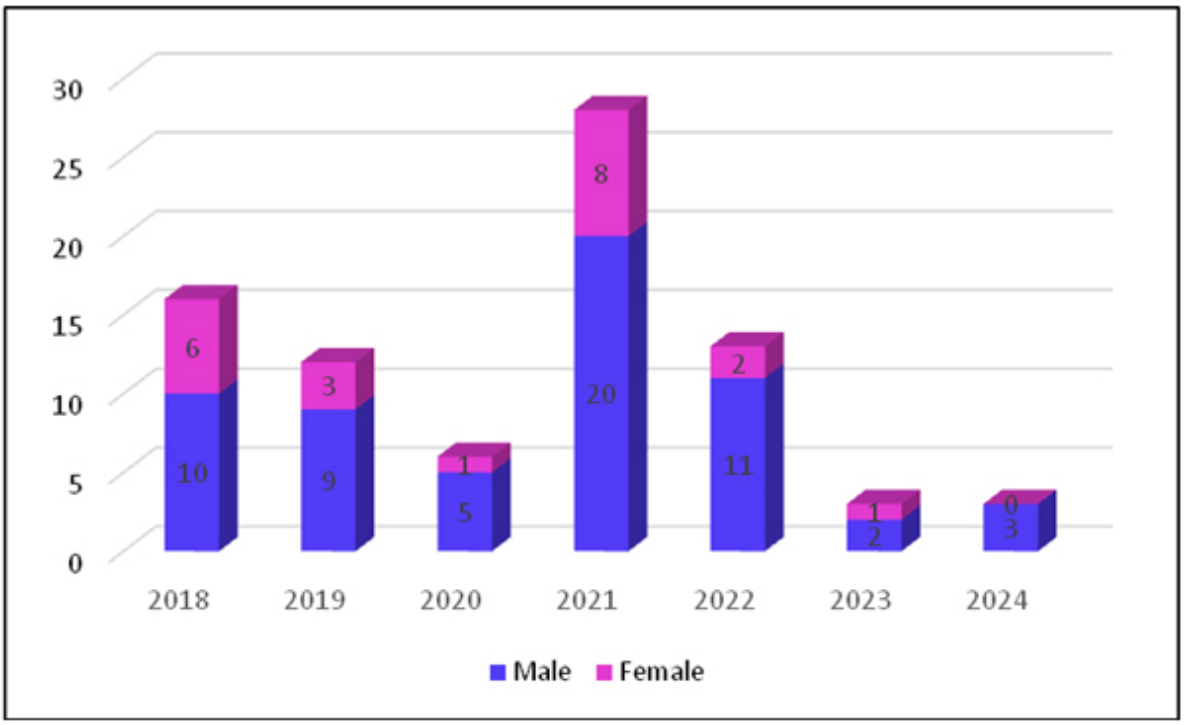

Figure 1: *Distribution of male/female neck CL cases according to years*

Table 1. Distrubition of cinical forms of CL lesions

Clinical Forms of Lesions	n	%
Noduloulcerative	90	61,7
Nodule	41	28,0
Papule	15	10,3
Total	146	100,0

Table 2. Distrubition of number of lesions according to age groups

	Number of Lesions									
Age Groups	1 lesion		2 lesions		3 lesions		≥4 lesions		Total	
1-18 years old	33	40,7	12	14,8	4	4,9	2	2,5	51	63,0
19-36 years old	7	8,6	3	3,7	0	0,0	3	3,7	13	16,0
37-54 years old	10	12,3	1	1,2	2	2,5	3	3,7	16	19,8
55 years old and older	1	1,2	0	0,0	0	0,0	0	0,0	1	1,2
Total	51	63,0	16	19,8	6	7,4	8	9,9	81	100,0

Table 3. The mean number, the average size and duration of lesions

The mean number of lesions		The average lesion size (mm)		The average lesion duration (month)	
Mean±SD	Range	Mean±SD	Range	Mean±SD	Range
1,8±1,5	1-9	14,38±3,05	3-60	11,56±0,79	4-40

DISCUSSION

Although no prior studies have exclusively focused on neck involvement in CL, a limited number of studies have specifically evaluated head and neck involvement in CL, where these regions are highly affected. In the study by Dunya et al., an analysis of the anatomical distribution of lesions in patients with head and neck involvement found that neck involvement accounted for only 3.1% of cases[1]. Studies reporting that when the head and neck regions are considered together, it is the region most commonly affected by CL. In Türkiye, studies have found that 60.7%, 58%, 57.3%, and 47.8% of patients with CL have head and neck region involvement, respectively. In the study of Uzun et al. who divided the head-neck region into detailed anatomical compartments and evaluated 1030 patients, only 1 lesion (0.1%) was found to be located in the neck, while in the study of Gurel et al. 2120 CL patients were evaluated and only 1.3% of the lesions were found to be located in the neck[7-10].

In a study conducted in Iran in children diagnosed with CL below the age of 13 years, neck involvement was found with a rate of 4.1%[11].

When the head and neck region is considered together in CL, it is the region most commonly affected by CL in most studies, but most of the involvement of this region is localized on the face, while neck involvement is either absent or very low. During the period of our study, there were a total of 3246 patients diagnosed with CL in the Oriental Boil Diagnosis and Treatment Center in Şanlıurfa province and the rate of patients with isolated neck involvement was 2.5%. This rate is higher than the rates in the studies of Uzun et al and Gurel et al, which were previously conducted in our country with more patients.

Since neck involvement in cutaneous leishmaniasis is rarely reported, our study will contribute to the literature in terms of the involvement of this anatomical region.

Cutaneous leishmaniasis is most commonly observed in pediatric age groups in endemic areas. The lower frequency of cases in adults may be due to acquired immunity following infection during childhood[10,12]. In alignment with the literature, the majority of patients in this study (63%) were aged 0-18 years.

In the study by Dunya et al., which compared patients with and without head and neck involvement, the frequency of head and neck involvement was significantly higher in patients under 18 years of age. The mean age of the group with head and neck involvement was significantly lower than that of the group without involvement. This was attributed to children's longer sleep duration and their increased exposure to vector bites while sleeping, particularly in the head and neck regions[1].

In the study by Gurel et al., 68.5% of patients had a single lesion, 17.8% had two lesions, 7.6% had three lesions, and 2.4% had four lesions, with a mean lesion count of 1.6[9]. In the study by Gurel et al., 68.5% of patients had a single lesion, 17.8% had two lesions, 7.6% had three lesions, and 2.4% had four lesions, with a mean lesion count of 1.8[12].

In this study, 63% of patients had a single lesion, while 37% had multiple lesions, with a mean lesion count of 1.8. These findings were consistent with previous studies.

In the study by İnci et al., the most common clinical lesion type was nodulo-ulcerative lesions (54%), followed by papulonodular lesions (43%)[13]. Similarly, in the study by Gurel et al., nodulo-ulcerative lesions were the most common type, observed in 39.2% of patients, followed by nodular lesions (31.4%) and papular lesions (16.6%)[9]. The distribution of lesion types in our study was similar to these studies conducted in Türkiye. The most common lesion type was nodulo-ulcerative lesions (61.7%), followed by nodular lesions (28.0%) and papular lesions (10.3%).

Cutaneous leishmaniasis is referred to as the "great imitator" because it can mimic most dermatoses. Living in endemic areas or recent travel to endemic regions, along with the presence of long-standing asymptomatic papules, nodules, plaques, ulcers, or crusted ulcers in uncovered body areas, should alert clinicians to

the possibility of CL. The diagnosis should be confirmed by parasitological or immunological laboratory methods[14,15].

As a first approach, it is recommended to obtain an appropriate sample for smearing, followed by staining with Giemsa and microscopic examination. Smear examination is a cost-effective, simple, and rapid diagnostic tool for CL. The detection of Leishmania amastigotes within and/or outside macrophages during microscopic examination, while not enabling species-level identification, is sufficient for diagnosis. A recently developed diagnostic algorithm suggests initiating Leishmania treatment if microscopic examination is positive with this method, and conducting further investigations via skin biopsy if results are negative. In our study, 75 patients (92.5%) were diagnosed by direct microscopic examination of affected skin, while in six patients (7.5%), the diagnosis was made via histopathological examination following a negative microscopic evaluation (Figure 2a-c). The diagnostic sensitivity of direct microscopic examination of cutaneous smear samples is significantly influenced by the examiner's experience[14,16]. All examinations in our study were conducted at the Diagnosis and Treatment Center for Oriental Boil, where the personnel have extensive experience with this method.

Once the diagnosis is confirmed, treatment is recommended to accelerate healing, reduce the likelihood of recurrence, prevent the formation of disfiguring scars and the risk of disease dissemination, and prevent the patient from serving as a reservoir for the disease[17]. For these reasons, we treated all our patients.

Treatment options for CL are classified as topical treatments, intralesional injections, physical therapies, and systemic therapies. Certain considerations must be taken into account when selecting a treatment. Systemic treatments are preferred for patients with multiple lesions (>5), large lesions (>5 cm), lesions in functionally sensitive areas, or mucosal involvement. In cases with single or a few small lesions, the most ideal treatments are physical, topical, or intralesional therapies. Intralesional antimonial therapy (meglumine antimonate and sodium stibogluconate) has been one of the best treatment options for many years. It is particularly considered the gold standard for treating solitary and small lesions[6]. The majority of our patients had solitary or a small number of small lesions. Therefore, intralesional meglumine antimonate (IL MA) therapy was administered to all except one patient with numerous and large lesions. Treatment was successful in all patients. The single patient with multiple and large lesions was treated with systemic MA therapy and achieved recovery.

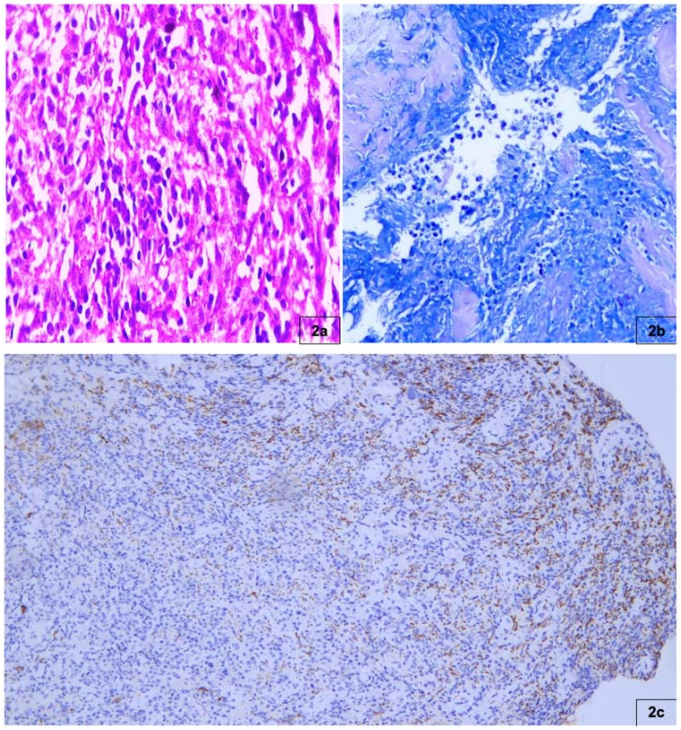

Figure 2a: *Leishmaniasis showing a mixed inflammatory cell infiltrate. The infiltrate includes amastogotes and lymphocytes(HE; x400) Figure 2b: Giemsa stain showing amastogotes of Leishmania. Figure 2c: CD1a positive amastigotes (x200)*

Limitations of Study

The retrospective nature of the study and the inability to perform species-level identification of the causative agent were the study's limitations.

CONCLUSION

In endemic areas, CL must be considered in the differential diagnosis of long-standing, non-healing lesions of various types on the neck. If suspected, laboratory investigations should confirm the diagnosis, and treatment should be initiated. IL MA therapy should be considered the first-line treatment for neck CL.

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