

CASE REPORT

DOI: 10.5336/caserep.2022-87971

Diagnosis and Treatment of Eyelid Cutaneous Leishmaniasis

^{id} Gizem DOĞAN GÖKÇE^a, ^{id} Sema İpek ALGAN^b, ^{id} İrem Şahver İŞGÖR^c, ^{id} Sacide ÇOLAK^d

^aClinic of Ophthalmology, Gönen State Hospital, Balıkesir, Türkiye

^bClinic Dermatology, Gönen State Hospital, Balıkesir, Türkiye

^cDepartment of Pathology, Ege University Faculty of Medicine, İzmir, Türkiye

^dClinic of Pathology, Balıkesir Atatürk City Hospital, Balıkesir, Türkiye

ABSTRACT Cutaneous leishmaniasis (CL) is the most common form of leishmaniasis transmitted by the Phlebotomus sand fly. The eyelid is rarely involved because of voluntary and involuntary blinking of the eyelid. Diagnosis of CL is often difficult due to its atypical location, misleading appearance and very long incubation period. When seen in non-endemic areas, the diagnosis of CL may not be considered firstly and it may cause delay in diagnosis and treatment. Although the disease usually regresses spontaneously, it may cause complications in cases of delayed treatment. In this study, we reported the pediatric patient with atipic appearance of ocular CL in a non-endemic area for leishmaniasis. We reported the diagnose and treatment processes of the patient with ocular CL by clinical, radiological and histopathological images.

Keywords: Eyelid infections; intralesional injection; leishmaniasis; meglumine antimonate

Cutaneous leishmaniasis (CL) is endemic in more than 90 countries worldwide.¹ World Health Organization states that there are 0.7-1 million cases of CL worldwide annually. Cases of 90% occur in Afghanistan, Pakistan, Syria, Saudi Arabia, Algeria, Iran, Brazil and Peru.² Due to migrations, the number of cases of CL has increased significantly, with imported cases being relatively common in non-endemic countries.³ CL is characterized by a single polymorphous lesion, usually located in the unclothed parts of body. In CL, the eyelid is rarely affected because of its movable structure and is seen in 0.4% of cases. CL can mimic basal cell carcinoma, hordeolum, chalazion and impetigo. It is important that when the diagnosis and treatment are delayed, the ocular process can progress to the ocular surface than retina and cause blindness.⁴

CASE REPORT

A 9-year-old male patient presented with painless redness and swelling on his right upper eyelid for 4

months. There was extensive hyperemia and papular lesion on the upper eyelid (Figure 1). There was no known additional systemic disease or drug use. In his complete ophthalmological examination, both visual levels were 0.0 according to LogMAR, eye movements were free in all directions.



FIGURE 1: The appearance of lesion on initial presentation. Approximately 3x1 cm erythematous plaque on the right upper eyelid and 0.5 cm diameter adherent crust on it.

Correspondence: Gizem DOĞAN GÖKÇE

Clinic of Ophthalmology, Gönen State Hospital, Balıkesir, Türkiye

E-mail: gizemdogann05@gmail.com



Peer review under responsibility of Türkiye Klinikleri Journal of Case Reports.

Received: 04 Jan 2022

Received in revised form: 18 Feb 2022

Accepted: 28 Feb 2022

Available online: 08 Mar 2022

2147-9291 / Copyright © 2022 by Türkiye Klinikleri. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

There was no accompanying ptosis and blepharitis, there were no conjunctival hyperemia and corneal infiltration, the other anterior segment structures and retina were normal in his slit-lamp examination. Blood tests were performed for infection differentiation. The blood results were in normal limits. Radiological imaging magnetic resonance was performed to evaluate the extent of the lesion and abscess formation (Figure 2). According to the information received from the patient's relatives, it was learned that they immigrated to Türkiye from Syria 1 year ago. Considering the lesion characteristics and the fact that the migrated region is among the risky areas for protozoan infections, CL with eyelid involvement was considered. For the definitive diagnosis, a 0.5 cm long, 2-3 mm deep incision was made on the edge of the lesion under sedation, and a biopsy was taken. At the same time, the biopsy material was imprinted on the slides and dried in air and evaluated by the pathologist by staining with May Grunwald Giemsa dye (Merck, Darmstadt, Germany). The diagnosis of leishmania was confirmed by the appearance of round shaped amastigote forms in pathology specimens (Figure 3). Although, different treatment modalities have been reported in ocular leishmaniasis cases, we planned to apply intralesional meglumine antimoniate injection. Considering the side effects of the drug before the treatment cardiac and systemic diseases were ruled out. We injected 1 mL meglumine antimoniate (Glucantime®, Tillotts Pharma AG, Switzerland) per week for 6 weeks without treatment anesthesia until whiteness was seen from the edges of the lesion to the middle. At the end of 6 weeks, the treatment was terminated as the lesion regressed with scarring. No progression was observed in the lesion in the 6-month follow-up. No additional skin lesions and injection related complications were developed. The lesion was recorded weekly with photographs and regression was demonstrated (Figure 4). Written consent was obtained from the patient's parents for case report.

DISCUSSION

Sand flies have short mouthparts, so they can not bite through clothing, and CL usually occurs on an exposed part of the body.⁵ Although the face is an un-

clothed area, it is an unusual area, especially for the eyelids.⁶ In addition, atypical clinical presentation or CL lesions in non-endemic areas may cause diagnostic difficulties. In our case report, we presented a pediatric patient with ocular CL from Türkiye and the Balıkesir region, which is not endemic for the disease. The other study from Türkiye about CL patient in the Şanlıurfa region, stated that 3.57% of the patients had eyelid involvement. They reported that mechanical ptosis developed in one patient with ocular involvement and lagophthalmos in 2 patients. The size of the lesions and their proximity to the valve marginal edge are important in the development of ocular signs.⁷ Although CL is a self-limiting disease, untreated ocular leishmaniasis can be a potential cause of blindness with corneal and retinal invasion.⁸ Initially, it appears as a polymorphous lesion in the form of swelling, papules, and plaques.⁹

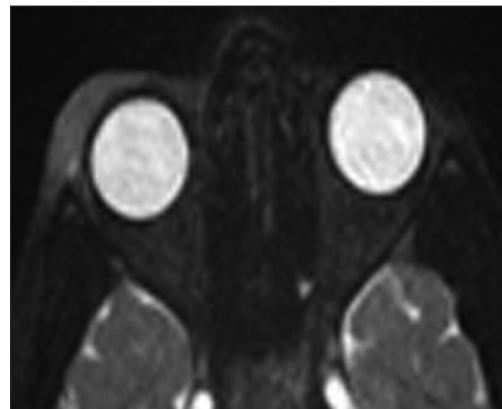


FIGURE 2: Cross-sectional image of the magnetic resonance T2 sequence. There is a hyperintense appearance of the skin and subcutaneous tissues on the right upper eyelid. No abscess formation or space-occupying lesions were seen.

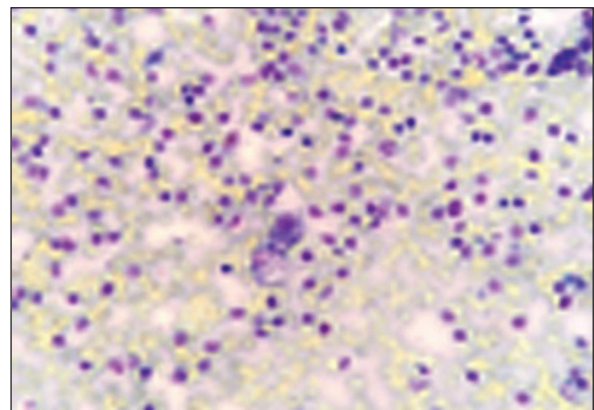


FIGURE 3: Amastigote forms show on May Grunwald Giemsa slides (400x).



FIGURE 4: A) Appearance of the lesion one week after the first injection. There was no significant regression in the lesion; B) Appearance of the lesion one week after the second injection. The erythema began to fade and the crust began to shrink; C) Appearance of the lesion one week after the third injection. There was significant regression in the induration of the lesion; D) Appearance of the lesion one week after the fourth injection; E) Appearance of the lesion one week after the fifth injection; F) Appearance of the lesion one week after the sixth injection. The lesion completely regressed, leaving an atrophic and erythematous scar.

In one study, several cases of eyelid CL with the appearance of nodules covered with crusting and crusting were reported.¹⁰ Because of this atypical appearance, eyelid CL maquerates different ophthalmologic conditions such as chalazion, basal cell carcinoma, infundibular cyst.¹¹⁻¹³ There is still no vaccine to prevent the disease, and the classic therapy for all forms of leishmaniasis are pentavalent antimoniate compound.¹⁴ In CL with adnexal involvement, systemic meglumine antimoniate (Glucantime®) (20 mg/kg for at least 3 weeks), intralesional meglumine antimoniate (Glucantime®) (1 mL/lesion per week for 2 weeks) twice, once a week for at least 2 months) or paromomycin ointment 20% (for at least 3 weeks for once- daily application) treatment protocols are recommended.¹⁵

We preferred intralesional injection to reduce the side-effect profile in our patient. The compliance of our patient in the treatment application was very important in its applicability. Leishmaniasis may not be diagnosed patients with atypical presentations seen in non-endemic regions and may lead to results that do not require delayed treatment. It should be noted that as in our patient, signs can be seen months after the fly bite. We wanted to emphasize that it is important to take a biopsy in suspicious cases and start

treatment in the early period. Our study is important in terms of showing the feasibility of intralesional injection in the pediatric age group and that patients with atypical CL can be encountered in non-endemic regions.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Gizem Doğan Gökçe; **Design:** Sema İpek Algan; **Control/Supervision:** Gizem Doğan Gökçe, Sema İpek Algan; **Data Collection and/or Processing:** Gizem Doğan Gökçe; **Analysis and/or Interpretation:** Sacide Çolak; **Literature Review:** Sema İpek Algan; **Writing the Article:** Gizem Doğan Gökçe, İrem Şahver İşgör; **Critical Review:** Sema İpek Algan; **References and Fundings:** Sacide Çolak; **Materials:** Sacide Çolak.

REFERENCES

1. Handler MZ, Patel PA, Kapila R, Al-Qubati Y, Schwartz RA. Cutaneous and mucocutaneous leishmaniasis: differential diagnosis, diagnosis, histopathology, and management. *J Am Acad Dermatol.* 2015;73(6):911-26; 927-8. [[Crossref](#)] [[PubMed](#)]
2. Desjeux P. Leishmaniasis: current situation and new perspectives. *Comp Immunol Microbiol Infect Dis.* 2004;27(5):305-18. [[Crossref](#)] [[PubMed](#)]
3. Pavli A, Maltezou HC. Leishmaniasis, an emerging infection in travelers. *Int J Infect Dis.* 2010;14(12):e1032-9. [[Crossref](#)] [[PubMed](#)]
4. Mohammadpour I, Motazedian MH, Handjani F, Hatam GR. Cutaneous leishmaniasis of the eyelids: a case series with molecular identification and literature review. *Korean J Parasitol.* 2016;54(6):787-92. [[Crossref](#)] [[PubMed](#)] [[PMC](#)]
5. Sadeghian G, Nilfroushzadeh MA, Moradi SH, Hanjani SH. Ocular leishmaniasis: a case report. *Dermatol Online J.* 2005;11(2):19. [[Crossref](#)] [[PubMed](#)]
6. Ayatollahi J, Ayatollahi A, Shahcheraghi SH. Cutaneous leishmaniasis of the eyelid: a case report. *Case Rep Infect Dis.* 2013;2013:214297. [[Crossref](#)] [[PubMed](#)] [[PMC](#)]
7. Satici A, Gurler B, Aslan G, Ozturk I. Ocular involvement in cutaneous leishmaniasis four cases with blepharoconjunctivitis. *Eur J Epidemiol.* 2004;19(3):263-6. [[Crossref](#)] [[PubMed](#)]
8. Abrishami M, Soheilian M, Farahi A, Dowlati Y. Successful treatment of ocular leishmaniasis. *Eur J Dermatol.* 2002;12(1):88-9. [[PubMed](#)]
9. Durdu M, Gökçe S, Bagirova M, Yalaz M, Allahverdiyev AM, Uzun S. Periocular involvement in cutaneous leishmaniasis. *J Eur Acad Dermatol Venereol.* 2007;21(2):214-8. [[Crossref](#)] [[PubMed](#)]
10. Mencia-Gutiérrez E, Gutiérrez-Díaz E, Rodríguez-Peralto JL, Monsalve-Córdova J. Old world eyelid cutaneous leishmaniasis: a case report. *Dermatol Online J.* 2005;11(3):29. [[PubMed](#)]
11. Hanafi Y, Oubaaz A. Leishmaniose palpébrale simulant un chalazion: à propos d'un cas [Leishmaniasis of the eyelid masquerading as a chalazion: case report]. *J Fr Ophtalmol.* 2018;41(1):e31-e3. French. [[Crossref](#)] [[PubMed](#)]
12. Jaouni T, Deckel Y, Frenkel S, Ilisar M, Pe'er J. Cutaneous leishmaniasis of the eyelid masquerading as basal cell carcinoma. *Can J Ophthalmol.* 2009;44(5):e47. [[Crossref](#)] [[PubMed](#)]
13. Veraldi S, Bottini S, Currò N, Gianotti R. Leishmaniasis of the eyelid mimicking an infundibular cyst and review of the literature on ocular leishmaniasis. *Int J Infect Dis.* 2010;14 Suppl 3:e230-2. [[Crossref](#)] [[PubMed](#)]
14. Kumar R, Engwerda C. Vaccines to prevent leishmaniasis. *Clin Transl Immunology.* 2014;3(3):e13. [[Crossref](#)] [[PubMed](#)] [[PMC](#)]
15. Faghihi G, Tavakoli-kia R. Treatment of cutaneous leishmaniasis with either topical paromomycin or intralesional meglumine antimoniate. *Clin Exp Dermatol.* 2003;28(1):13-6. [[Crossref](#)] [[PubMed](#)]