Letter to the Editor
Editöre Mektup
DOI: 10.4274/turkderm.galenos.2019.79477
Turkderm - Turk Arch Dermatol Venereol 2020;54:36-7

A neglected disease in a patient with dermatomyositis: Cutaneous leishmaniasis

Bir dermatomiyozit hastasında ihmal edilmiş bir hastalık: Kutan leşmanyazis

Özlem Özbağçıvan, Sevgi Akarsu, Şebnem Aktan, Banu Lebe*, Emel Fetil
Dokuz Eylül University Faculty of Medicine, Department of Dermatology;
*Department of Pathology, İzmir, Turkey

Keywords: Cutaneous, leishmaniasis, dermatomyosisit, immunosupresion

Anahtar Kelimeler: Kutan, leşmanyazis, dermatomiyozit, immünosupresyon

To The Editor,

A 66 year old woman presented with a 1,5-year history of painless ulcer on the ankle. The lesion had started as a single small papule, and then gradually increased in size despite topical and systemic antibiotic treatment. She described no relevant trauma or infection history, and there were no other similar appearing lesions elsewhere on the body. Past medical history was significant for dermatomyositis for 8 years, and she was treated with azathioprine for more than 6 years. On physical examination, there was an ulceration of 4x3 cm size with an erythematous border (Figure 1). Laboratory analysis revealed a normal white blood cell count of 7.900/mm3 with 92.3% segmented neutrophils, 5.7% monocytes, and 4.2% lymphocytes, C-reactive protein level of 10 mg/L (0.2 to 5), and erythrocyte sedimentation rate level of 81 mm/h (0 to 20). Serological investigations, including those for human immunodeficiency virus, syphilis, hepatitis B and C virus yielded negative. Bacteriological and mycological cultures taken from the lesion were negative. A further skin biopsy was taken from the lesion with preliminary diagnosis of pyoderma gangrenosum, vasculopathic or vasculitic ulcers associated with dermatomyositis, or atypical mycobacterial

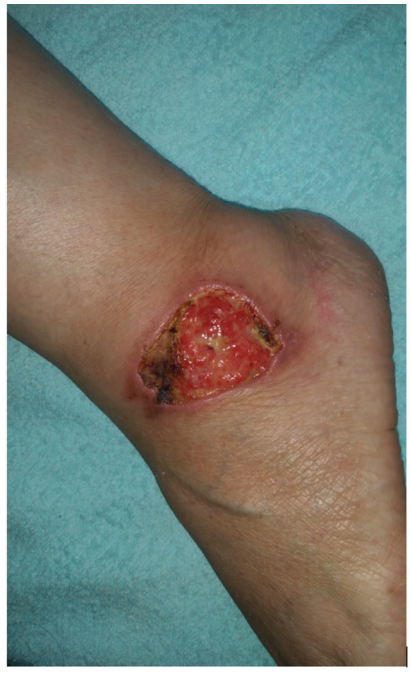

Figure 1. Ulceration with an erythematous border on the ankle

Address for Correspondence/Yazışma Adresi: Özlem Özbağçıvan MD, Dokuz Eylül University Faculty of Medicine, Department of Dermatology, İzmir, Turkey
Phone: +90 505 295 25 66 E-mail: ozlem.ozbagcivan@deu.edu.tr **Received/Geliş Tarihi:** 20.03.2019 **Accepted/Kabul Tarihi:**11.12.2019
ORCID: orcid.org/0000-0001-7190-3969

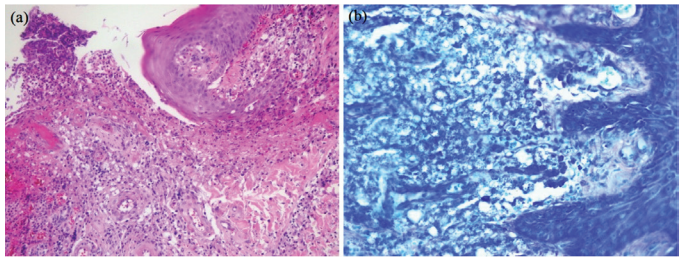

Figure 2. a) Dense superficial dermal infiltrate composed of lymphocytes and histiocytes filled with numerous small intracytoplasmic structures, b) Leishmania amastigotes stained positive with Giemsa-stain in the cytoplasm of histiocytes

infections; but histological examination found a dense superficial dermal infiltrate composed of lymphocytes and histiocytes filled with numerous small intracytoplasmic structures, suggesting leishmaniasis in hematoxylin-eosin staining (Figure 2a), and in Giemsa staining (Figure 2b). Microscopic examination of Giemsa-stained slides of scrapings from the lesion also revealed the amastigotes. The patient then treated with systemic antimony (Glucantime) with complete healing.

Cutaneous leishmaniasis (CL), caused by a parasitic infection, appears to be increasing in worldwide because of extended urbanization. It can be caused by several Leishmania spp. and is transmitted to human beings and animals by sandflies. The diagnosis may sometimes be challenging because of the pleomorphic clinical manifestations, and can be indistinguishable from lesions related to other diseases[1,2]. Despite its increasing incidence, CL is considered one of the most overlooked diseases worldwide[3].

Immunosuppression is a well-established risk factor for CL. Clinical presentation can be atypical in immunosuppressed patients, being easily misdiagnosed as other dermatological conditions. Additionally, when the patient had also a previously known systemic disease, CL can be easily neglected with the possibility of a condition related to the previous disease, treatment, or a flare-up of the underlying condition. However, a delay in diagnosis in immunosuppressed patients may lead to an unfavorable outcome, especially if severe, including parasite dissemination, visceralization, and even death[4].

The diagnosis of CL is often made clinically and, if possible, by microscopic examination of lesion biopsy and smears to visually confirm the parasites as the cause[5]. Although numerous local and systemic treatments are available for leishmaniasis, the mainstays of CL treatment are pentavalent antimonials which have been used successfully worldwide[1].

Given the presence of dermatomyositis and immunosuppresive therapy in her past medical history, we primarily considered pyoderma gangrenosum, vasculopathic or vasculitic ulcers associated with dermatomyositis, bacterial, deep fungal or atypical mycobacterial infections in the clinical differential diagnosis of the patient. Due to the rarity of cases in our region, CL was not the first suspected diagnosis until the patient recalled that she had travelled for a short trip to Eastern Anatolia two years ago, where CL is endemic.

To the best of our knowledge, there is no such report in the existing literature describing the coexistence of dermatomyositis and CL. This case has been of clinical interest to emphasize that CL may associate with other systemic dermatological diseases, and clinicians should be aware of sporadic CL cases even in non-endemic areas. Informed consent wasn't obtained.

Ethics

Informed Consent: It wasn't obtained.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: Ö.Ö., S.A., Ş.A., B.L., E.F., Concept: Ö.Ö., Design: Ö.Ö., Data Collection or Processing: Ö.Ö., B.L., Analysis or Interpretation: Ö.Ö., S.A., Ş.A., B.L., E.F., Literature Search: Ö.Ö., Writing: Ö.Ö.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Aytekin S: Treatment approaches for cutaneous leishmaniasis. Turkderm 2009;43:44-7.
2. Ceyhan AM, Meric G, Aynali G: A Case of cutaneous leishmaniasis mimicking squamous cell carcinoma. Turkderm 2012;46:44-6.
3. Aronson NE, Joya CA: Cutaneous leishmaniasis: updates in diagnosis and management. Infect Dis Clin North Am 2019;33:101-117.
4. Juzlova K, Votrubova J, Kacerovska D, et al: Visceral leishmaniasis with cutaneous symptoms in a patient treated with infliximab followed by fatal consequences. Dermatol Ther 2014;27:131-4.
5. Burza S, Croft SL, Boelaert M: Leishmaniasis. Lancet 2018;392:951-70.