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A Case of Tumid Lupus Erythematosus Successfully Treated with Topical Tacrolimus and Hydrocortisone 17-Butyrate

Topikal Takrolimus ve Hidrokortizon 17-Butirat ile Başarıyla Tedavi Edilen Bir Tumid Lupus Eritematozus Olgusu

Funda Tamer[1], İrem Özdemir[1], Betül Öğüt[2], Özlem Erdem[2]

[1]Department of Dermatology, Gazi University Faculty of Medicine, Ankara, Türkiye
[2]Department of Pathology, Gazi University Faculty of Medicine, Ankara, Türkiye

ABSTRACT

Tumid lupus erythematosus is an uncommon subtype of chronic cutaneous lupus erythematosus. Hereby, we report a 36-year-old male patient with tumid lupus erythematosus who presented with erythematous papules and plaques on the face and neck who showed significant improvement after treatment with topical tacrolimus and topical hydrocortisone 17-butyrate.

Keywords: Cutaneous lupus erythematosus, treatment, tumid lupus erythematosus

ÖZ

Tumid lupus eritematozus, kronik kutanöz lupus eritematozusun nadir görülen bir alt tipidir. Burada, yüz ve boyunda eritemli papül ve plaklarla başvuran ve topikal takrolimus ve topikal hidrokortizon 17-butirat tedavisi sonrasında belirgin iyileşme gösteren, tumid lupus eritematozuslu 36 yaşında bir erkek hastayı sunduk.

Anahtar Sözcükler: Kutanöz lupus eritematozus, tedavi, tumid lupus eritematozus

INTRODUCTION

Tumid lupus erythematosus is an uncommon subtype of chronic cutaneous lupus erythematosus. However, it has been suggested that tumid lupus erythematosus might be considered as a distinct entity since the relationship between the disease and systemic lupus erythematosus and serological abnormalities were scarce (1,2). Tumid lupus erythematosus affects females and males equally. The disease usually occurs in individuals aged 30-40 years. Nevertheless, the incidence and prevalence of this disease among different races and ethnicities remain unknown (2). Tumid lupus erythematosus is characterized by erythematous or violaceous annular plaques with edema in sun-exposed areas (2,3). The face, neck, chest, and back are the most commonly affected body areas. Various skin diseases, such as polymorphic light eruption, pseudolymphoma of the skin, and reticular erythematous mucinosis, should be included in the differential diagnosis of tumid lupus erythematosus. Topical and intralesional steroids, calcineurin inhibitors, antimalarial drugs, methotrexate, and mycophenolate mofetil are the treatment options (2). Patients with tumid lupus erythematosus should also be recommended to apply sunscreen regularly.

CASE REPORT

A 36-year-old male patient was admitted with a 6-year history of asymptomatic, erythematous plaques on the face. The lesions appeared on the cheek and gradually distributed to the face and neck. In addition, the patient stated that the lesions were exacerbated by ultraviolet radiation and heat exposure. Nevertheless, the patient reported no daily use of sunscreen. The patient was admitted to

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Address for Correspondence/Yazışma Adresi: Funda Tamer MD, Department of Dermatology, Gazi University School of Medicine, Ankara, Türkiye
E-mail / E-posta: fundatmr@yahoo.com
ORCID ID: orcid.org/0000-0003-4157-5583

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a dermatology clinic for skin lesion treatment a year ago. He was recommended to use 1% pimecrolimus cream and 0.05% clobetasol 17-propionate ointment twice daily for three months. However, the patient stated that he did not apply adequate treatment, and no clinical response was achieved. The past medical history was remarkable for hypertension and liver enzyme elevation associated with alcohol intake. However, regular use of any medication was denied. The family history was unremarkable. Dermatological examination revealed erythematous, squamous papules, and plaques on the face and neck (Figure 1a-c).

A skin biopsy was performed from an erythematous plaque on the patient's face to reach a definitive diagnosis. Histopathological examination revealed a normal epidermis, perivascular mononuclear inflammation involving plasma cells and lymphocytes, and mucin deposition in areas containing perivascular inflammation (Figure 2). Thus, the diagnosis of tumid lupus erythematosus was made based on clinical and histopathological findings. Among the laboratory tests, the biochemistry panel revealed increased levels of alanine aminotransferase (90 U/L, normal limit: <49 U/L), aspartate aminotransferase (60 U/L, normal limit: <34 U/L), triglyceride (210 mg/dL, range: 0-150 mg/dL) and decreased level of high-density lipoprotein cholesterol (27.1 mg/dL, normal limit: >49 mg/dL). Complete blood count revealed increased white blood cell count (12.2 $x10^3$/µL, range: 3.91-10.9 $x10^3$/µL) and absolute neutrophil count (8.2 $x10^3$/µL, range: 1.8-6.98 $x10^3$/µL). Complement C3 and C4 levels were within normal limits. The serum antinuclear antibody was negative, whereas the anti-SSA (anti-Ro) antibodies were positive. Anti-double-stranded (ds) DNA, anti-Smith (Sm), anti-SSB (anti-La), antineutrophil cytoplasmic antibodies, anticardiolipin IgM, and anticardiolipin IgG were also negative. The patient received 0.1% tacrolimus ointment and 0.1% hydrocortisone 17-butyrate cream twice daily. Moreover, the patient was recommended to regularly use a sunscreen with a sun protection factor of 50. A good clinical response was achieved four weeks after treatment; however, the lesions left hypopigmentation (Figure 1d-f). The patient gave written informed consent to publish the case details. The patient gave written informed consent to publish the case details.

DISCUSSION

Tumid lupus erythematosus is a rare type of chronic cutaneous lupus erythematosus that was first described in 1909. The diagnosis of tumid lupus erythematosus is not always easy to make since the disease is rare and shows different clinical and histopathological features compared with other chronic cutaneous lupus erythematosus lesions (1). Moreover, the association between tumid lupus erythematosus and systemic lupus erythematosus is uncommon. The disease presents with erythematous, indurated, annular papules, and plaques on the face and trunk exacerbated by sun exposure (2). Scleredema-like presentation and unilateral blaschkolinear distribution have also been reported in patients with tumid lupus erythematosus (3,4). The lesions heal without leaving scars or pigmentary changes (5,6). Antinuclear antibodies or anti-

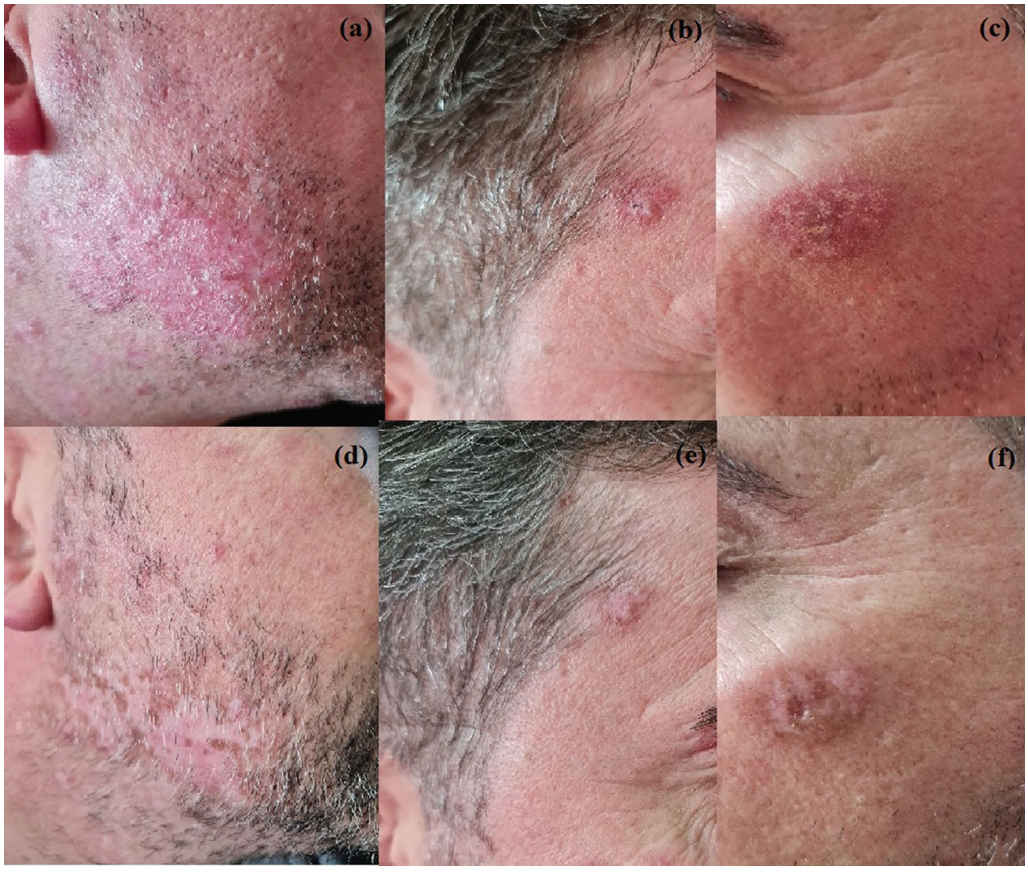

Figure 1. (a-c) Erythematous squamous papules, and plaques on the face and neck before treatment. (d-f) The lesions left hypopigmentation four weeks after treatment

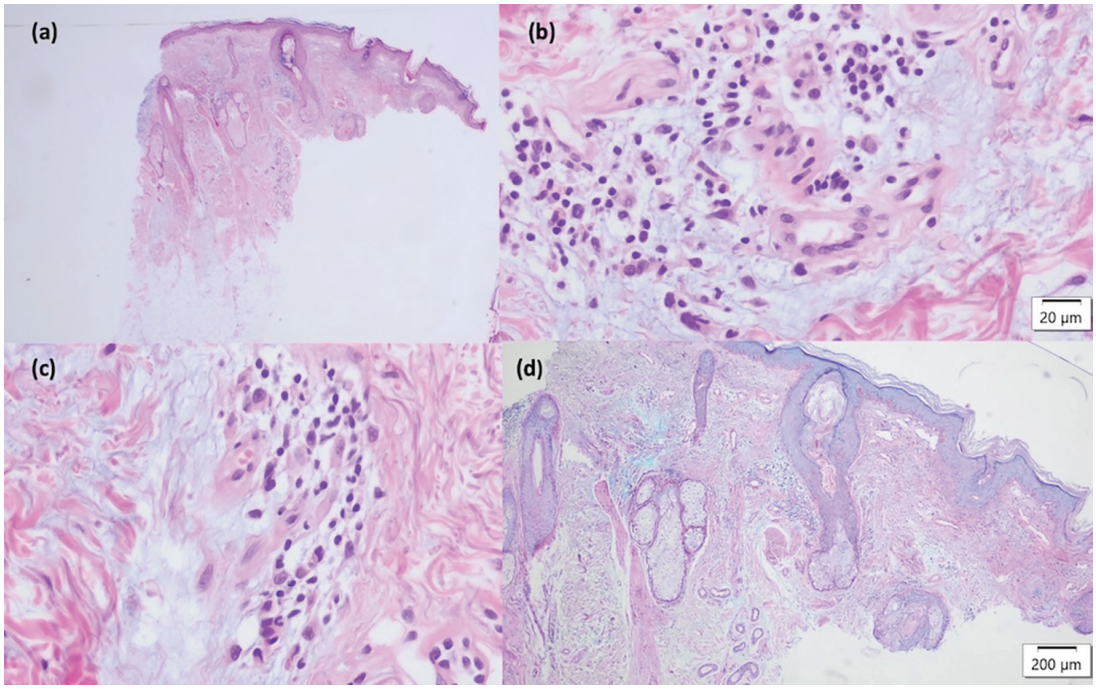

Figure 2. Histopathological examination revealed (a) Normal epidermis and skin appendages (Hematoxylin and eosin, x12.5). (b,c) Perivascular mononuclear inflammation involved plasma cells and lymphocytes (Hematoxylin and eosin, x400). (d) Mucin deposition in areas contained perivascular inflammation (Alcian blue, x200)

Ro/SSA, anti-La/SSB, anti-Sm, and anti-dsDNA antibodies are not detected in most patients. It has been suggested that sun protection and the use of topical corticosteroids and systemic antimalarial drugs might be effective for the treatment of tumid lupus erythematosus (6).

CONCLUSION

Herein, we present a rare case of tumid lupus erythematosus with erythematous, squamous papules, and plaques on the face and neck that rapidly responded to topical steroid and tacrolimus treatment. The presence of anti-Ro antibodies and lesions healed by leaving hypopigmentation, which were detected in our patient, were also uncommon findings in tumid lupus erythematosus. Our case will hopefully contribute to the literature by identifying the clinical features of the disease, improving diagnostic rates, and establishing the most appropriate treatment options.

Ethics

Informed Consent: The patient gave written informed consent to publish the case details.

Footnotes

Authorship Contributions

Surgical and Medical Practices: F.T., Concept: F.T., Design: F.T., Supervision: F.T., Ö.E., Resources: F.T., Material: F.T., B.Ö., Ö.E., Data Collection or Processing: F.T., İ.Ö., B.Ö., Ö.E., Analysis or Interpretation: F.T., B.Ö., Ö.E., Literature Search: F.T., Writing: F.T., İ.Ö., B.Ö., Ö.E., Critical Review: F.T., İ.Ö., B.Ö., Ö.E.

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

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