

SHORT COMMUNICATION

Lymphocytic Infiltration of the Eyelid: Two Cases Responding to Tacrolimus and Topical Glucocorticoid

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Jessner's lymphocytic infiltration of the skin (JLIS) is a skin disorder characterized by persistent papular and plaque-like eruptions (1). Although remission usually occurs spontaneously, JLIS has been known to have a high rate of relapse. The cause is unknown and can be hereditary. Rather than an independent disease, it has been suggested that JLIS may be a progressive stage of cutaneous lupus erythematosus or chronic primary T-cell lymphoma (2). Management can be performed in many different ways. We describe here 2 cases with lymphocytic infiltrate of the skin, which underwent complete regression after treatment with topical glucocorticoid or tacrolimus.

CASE REPORTS

Case 1. A 45-year-old woman had developed a solitary pea-like erythematous papular lesion on the left upper eyelid 2 months previously, which, although it had developed rapidly to a size of 30 × 35 mm (Fig. 1a) over a period of a few days, was completely asymptomatic in all other regards. Physical examination demonstrated a diffused tumid erythema on the left eyelid, which was soft on palpation. The patient had no history of any other disease. There was no evidence of systemic involvement. No lymphadenectasis had been detected. Serological studies showed no abnormality, other than being positive for anti-nuclear antibody, with a titre of 1:1000. Histopathology revealed regular epidermis, with a dense perivascular and periaxonal infiltration of lymphocytes in the dermis (Fig. 1c). PCR analysis showed no significant lymphocyte gene rearrangement. Immunohistochemical examination revealed CD3, CD3ε T cells

(Fig. 1e, f) assembled in the dermis, negative CD20, CD79, along with less than 5% positive Ki-67 (Fig. 1d). On the basis of these examinations, the diagnosis of Jessner's lymphocytic infiltrate of the skin was confirmed.

Treatment was commenced with topical applied tacrolimus ointment twice a day. The lesion began to recess after 2 days, and in the following 2 weeks the lesion gradually regressed to a pink macule (Fig. 1b). The patient remained symptom-free during the course of treatment. There was no recurrence in more than 5 months of follow-up.

Case 2. A 24-year-old woman presented with oedematous erythematous plaque on the right eyelid (Fig. 2a) with pruritus for 2 months. During this time, the lesion gradually progressed to reddish and expanded without effusion. No evidence was found that the lesions resulted from photosensitivity. Physical examination showed a scaly erythematous infiltrated plaque that involved only her right eyelid. In addition, oedema of the cheeks and swollen submaxillary lymph nodes was observed. Serological examinations showed leukocytopenia and neutropenia, antinuclear antibodies remained negative, but immunoglobulin E (IgE) was slightly increased. Lymphadenectasis of the neck and parotid were revealed by colour Doppler ultrasound. Histological examination revealed a dense lymphocytic infiltration in the dermis with no change in the epidermis. Immunohistochemistry staining revealed lymphocytic aggregates for CD3⁺, CD8⁺, CD2⁺, and CD5⁺ T cells (Fig. S1a–d; available from: <http://www.medicaljournals.se/acta/content/?doi=10.2340/00015555-1554>) in the dermis, in addition to a rate of Ki-67 less than 10% (Fig. S1e). Moreover, suspicious gene-rearrangement of T-cell receptor- γ (TCR- γ) was not observed. In short, the pathological and clinical findings suggested LIS. Treatment with mometasone furoate ointment once daily was commenced. Limited relief from the lesions was obtained in the following 5 days. However, a relapse, manifesting as diffuse

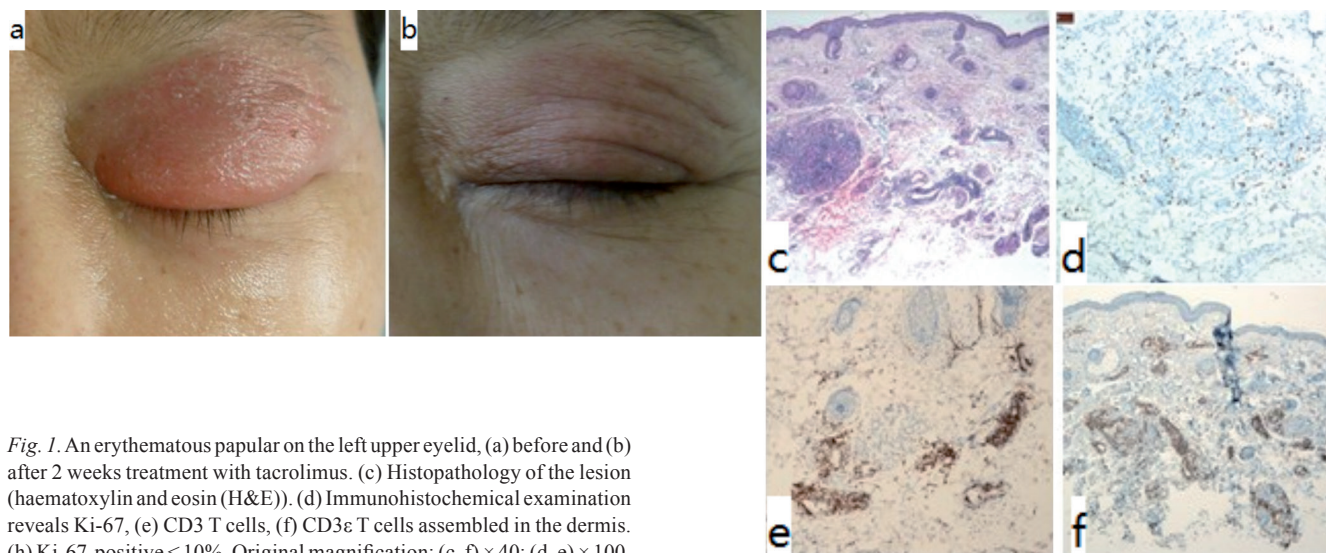


Fig. 1. An erythematous papular on the left upper eyelid, (a) before and (b) after 2 weeks treatment with tacrolimus. (c) Histopathology of the lesion (haematoxylin and eosin (H&E)). (d) Immunohistochemical examination reveals Ki-67, (e) CD3 T cells, (f) CD3ε T cells assembled in the dermis. (h) Ki-67-positive < 10%. Original magnification: (c, f) × 40; (d, e) × 100.

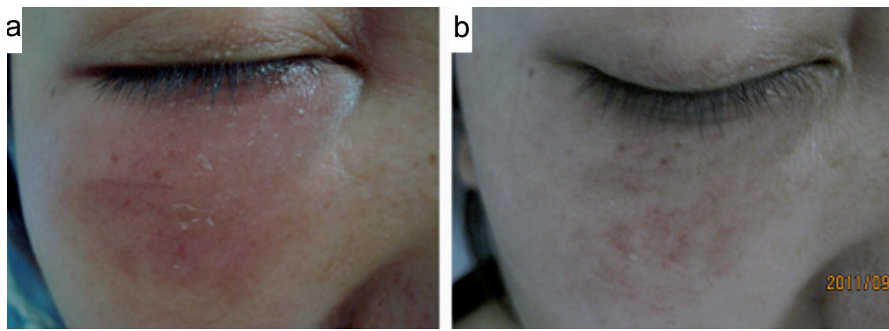


Fig. 2. An erythematous papule on the right eyelid (a) before and (b) after 2 weeks of treatment with mometasone furoate.

erythema and a tumid right eyelid, was noticed on the sixth day (Fig. 2b). We therefore changed treatment to mometasone furoate ointment twice daily. Complete regression of the lesion was attained after 2 weeks; and no lymph nodes were palpable. The patient had no recurrence during one year of follow-up.

DISCUSSION

The aetio-pathogenesis of JLIS is yet unclear; it is debatable whether it is a single entity or a heterogeneous group that can alter to polymorphous light eruption, discoid lupus erythematosus, or even malignant lymphoma (3, 4). Plenty of researchers have indicated that JLIS should not serve as an independent entity, but as a variant of cutaneous lupus erythematosus; although evidence to support this hypothesis is lacking (5). In addition, in some cases, lymphocytic infiltration of the skin and polymorphous light eruption can occur simultaneously. It is reported that JLIS occasionally represents clinical manifestations of *Borrelia*-associated pseudolymphoma (6). Typically, the lesions are flat discoid, elevated, and pinkish to reddish-brown, starting as diffuse papules; there may be only one or numerous lesions. JLIS presents mostly on the face, but also on sites exposed to the sun, such as the neck, upper trunk or proximal limbs of middle-aged adults. Interestingly, these 2 cases show certain typical and similar sites, eyelid and a part of the cheek, the only dissimilarity being the difference between the right and left sides. We did include the possibility of sun-exposure; however detailed history-taking revealed no relevant sun exposure related issues. Histological examination revealed a dense lymphocytic infiltrate in the dermis with no modification of the epidermis. The distinguishing features of JLIS from polymorphous light eruption and discoid lupus erythematosus are suppressor CD8⁺ T infiltration lymphocytes and absent dermal mucin and oedema.

Management options are diverse; most are useful, but patients do not respond to all options in the same way. Tacrolimus is an immunosuppressive drug that is used mainly to reduce the activity of the patient's immune system. Tacrolimus is also used as a topical preparation in the treatment of variety of skin conditions. Topical

and systemic glucocorticoid is traditionally used, but is not very effective in most cases. The first patient was treated successfully with topical tacrolimus ointment. The second patient was managed satisfactorily with a simple topical glucocorticoid ointment. Immunosuppressors, such as methotrexate, have been applied successfully in LIS; the antimalarial always work on some of those photosensitive cases (7). However, for some persistent and refractory LIS lesions, radiotherapy can be used; pulsed-dye laser and photodynamic therapy have also been reported recently (8). In the cases described here, topical tacrolimus exerted a therapeutic effect. Further long-term study will determine whether there have been any relapses or deterioration.

The authors declare no conflicts of interest.

REFERENCES

1. Calnan CD. Lymphocytic infiltration of the skin (Jessner). *Br J Dermatol* 1957; 69: 169–173.
2. Willemze R, Dijkstra A, Meijer CJLM. Lymphocytic infiltration of the skin (Jessner): a T-cell lymphoproliferative disease. *Br J Dermatol* 1984; 110: 523–529.
3. Willemze R, Vermeer BJ, Meijer CJLM. Immunohistochemical studies in lymphocytic infiltration of the skin (Jessner) and discoid lupus erythematosus: a comparative study. *J Am Acad Dermatol* 1984; 11: 832–840.
4. Rémy-Leroux V, Léonard F, Lambert D, Wechsler J, Cribier B, Thomas P, et al. Comparison of histopathologic–clinical characteristics of Jessner's lymphocytic infiltration of the skin and lupus erythematosus tumidus: multicenter study of 46 cases. *J Am Acad Dermatol* 2008; 58: 217–223.
5. Toonstra J, Wildschut A, Boer J, Smeenk G, Willemze R, van der Putte SC, et al. Jessner's lymphocytic infiltration of the skin: a clinical study of 100 patients. *Arch Dermatol* 1989; 125: 1525–1530.
6. Ziemer M, Eisendle K, Müller H, Zelger B. Lymphocytic infiltration of the skin (Jessner–Kanof) but not reticular erythematous mucinosis occasionally represents clinical manifestations of *Borrelia*-associated pseudolymphoma. *Br J Dermatol* 2009; 161: 583–590.
7. Guillaume JC, Moulin G, Dieng MT, Poli F, Morel P, Souteyrand P, et al. Crossover study of thalidomide vs placebo in Jessner's lymphocytic infiltration of the skin. *Arch Dermatol* 1995; 131: 1032–1035.
8. Borges da Costa J, Boixeda P, Moreno C. Pulsed-dye laser treatment of Jessner lymphocytic infiltration of the skin. *J Eur Acad Dermatol Venereol* 2009; 23: 595–596.