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Simultaneous occurrence of pemphigus foliaceus and bullous pemphigoid with concomitant herpesvirus infection

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Pemphigus foliaceus (PF) and bullous pemphigoid (BP), two distinct entities of autoimmune blistering diseases, rarely coexist. We report a patient who simultaneously developed both conditions in conjunction with a disseminated herpesvirus infection.

A dermatology consultation was requested for a 71-year-old man (Fig. 1a, b), who had been admitted to hospital for pneumonia with respiratory failure. A physical examination found generalized skin lesions on the patient's body; the oral and genital mucous membranes were spared.

Histological examination of a skin biopsy taken across the border of the erosion was performed (Fig. 2a–c), which found acantholysis in the epidermal granular layer, with oedema perivascular and interstitial infiltration of lymphocytes and eosinophils in the dermis. Direct immunofluorescence showed deposition of linear IgG at both the dermoepidermal junction and in the intercellular regions of the epidermis (Fig. 2d). Indirect immunofluorescence performed on human salt-split skin using the patient's sera found linear IgG deposition on the epidermal side and in the intercellular region between keratinocytes, both at a titre of 1 : 160. The antigenic specificities of the circulating antibodies, reacting with desmoglein1 and BP230 protein, were confirmed by Western blotting using human skin lysate as the substrate. Based on these findings, a diagnosis of coexisting PF and BP was made.

The patient was initially treated with systemic prednisolone at a dose of 60 mg daily but showed only partial improvement. On re-examination of the microscopic slides, scattered multinucleated giant cells, indicative of herpesvirus infection, were found on the eroded areas. A virus culture using a sample from the ruptured vesicles showed herpes simplex virus 2, and a disseminated herpesvirus infection was diagnosed. Consequently, a 7-day course of intravenous aciclovir was given, which produced additional improvement in the skin lesions. The dosage of prednisolone was gradually tapered to 20 mg daily, and azathioprine was added at a dose of 50 mg daily, as a steroid-sparing agent. In the following 2 years, recurrent episodes of herpesvirus infection occurred and each was well controlled by an oral antiviral agent without the need for tapering of the immunosuppressive drugs.

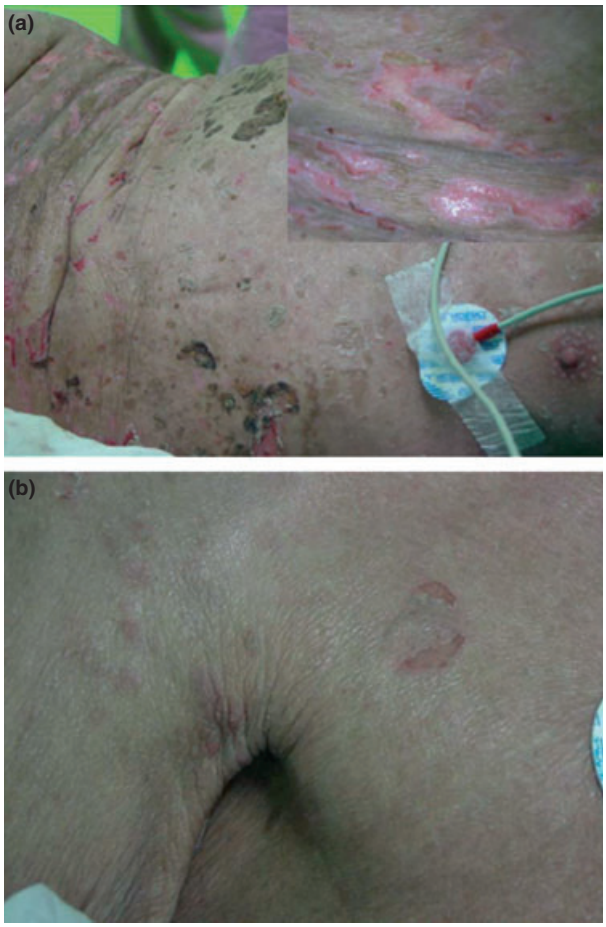

Figure 1 (a) Widespread erosions and crusts noted on the trunk. Similar lesions were also found on the face, neck and limbs. (b) Intact vesicles on an erythematous base found around the right axilla.

Coexistence of PF and BP has been reported in five previous cases, with two different types of presentation. Three of the five patients showed findings of both PF and BP but PF preceded or followed the development of BP by years.[1] The other two patients developed BP years after diagnosis of PF, but presented different clinical and

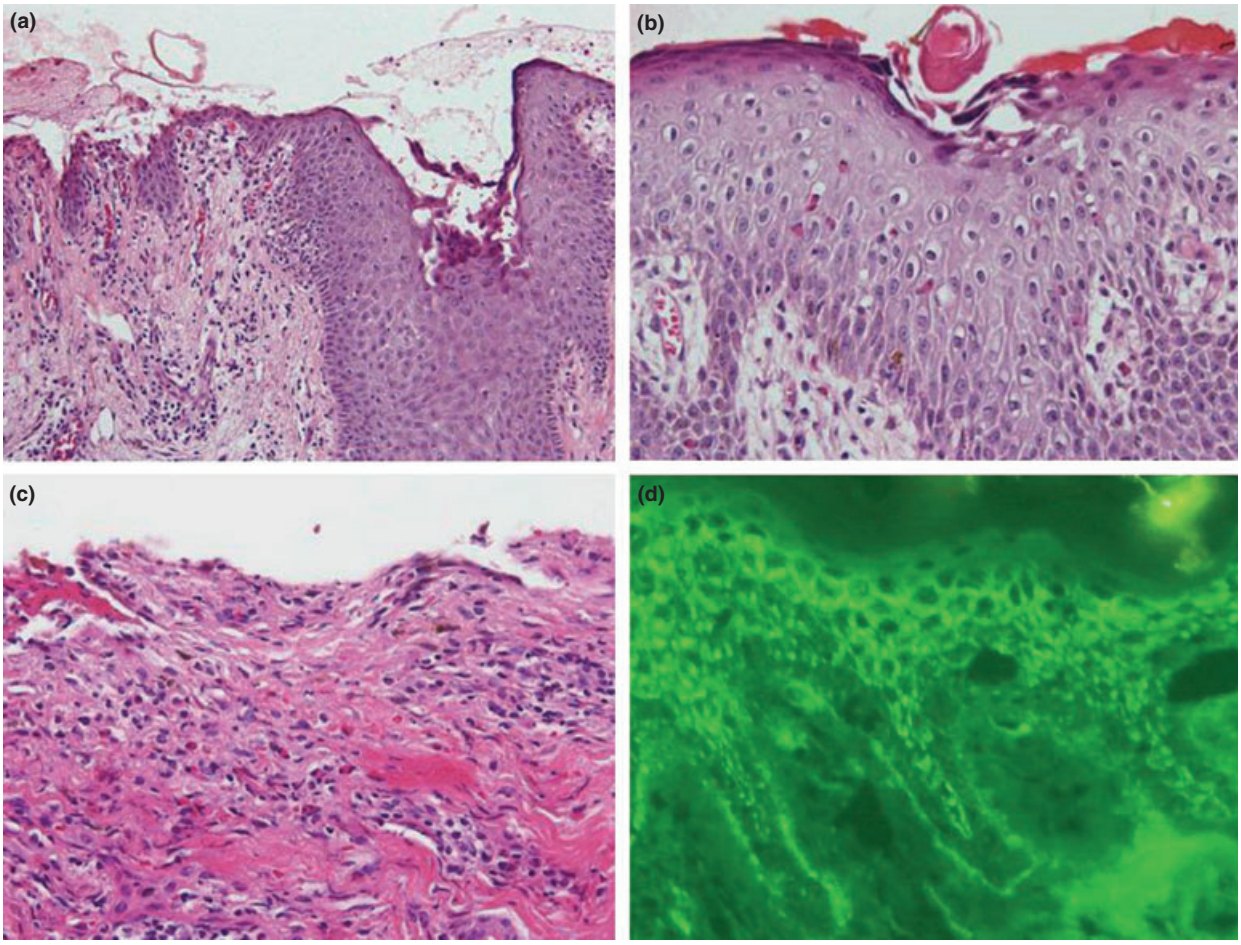

Figure 2 (a) Acantholysis noted in the epidermal granular layer, with oedema and perivascular and interstitial infiltration of lymphocytes and eosinophils in the dermis. (b) Acantholysis in the granular layer, with eosinophilic spongiosis. (c) Adjacent area showing loss of the epidermis; perivascular and interstitial infiltration of lymphocytes and eosinophils is present in the dermis. Haematoxylin and eosin; original magnification (a) × 100; (b,c) × 200) (d) Direct immunofluorescence showing deposition of linear IgG at both the dermoepidermal junction and intercellular regions of the epidermis (original magnification × 200).

laboratory findings at different timepoints, indicating a rare transitional form of PF to BP. The hypothesis of intermolecular epitope spreading was proposed to explain the shift of PF to BP or vice versa. Our patient is, to our knowledge, the first report of simultaneous occurrence of BP and PF, with both in the same lesion.

Clinicians should consider the possibility of herpesvirus infection in patients with pemphigus or pemphigoid, especially in those having a recalcitrant course. Hence, a high index of suspicion is needed when examining the histopathological slides of tissue from patients with bullous disorders.

This unusual scenario is a diagnostic and therapeutic challenge, and its possible pathogenetic significance is of interest. In such patients, early identification and treatment of concomitant herpetic infection is essential. In our experience, such herpesvirus infection can be well controlled by prompt introduction of antiviral agents without the need to taper systemic corticosteroids.

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Increased serum thymic stromal lymphopoietin levels in patients with cutaneous T cell lymphoma

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Thymic stromal lymphopoietin (TSLP) was first cloned from a murine thymic stromal cell line.[1] Human TSLP potently activates immature CD11c-positive myeloid dendritic cells (DCs), which create a T-helper (Th)-2-permissive microenviroment.[2,3] *In vivo*, TSLP has been shown to be strongly expressed by keratinocytes in acute and chronic lesions of atopic dermatitis (AD), but not expressed in normal skin or nonlesional skin in patients with AD.[2] There has been, however, no report studying serum TSLP levels in patients with skin diseases.

The medical ethics committee approved the study, and informed consent was obtained from all subjects. In total, 13 patients with AD (mean ± SD age 31.1 ± 14.5 years), 12 patients with psoriasis (56.9 ± 14.3 years), 29 patients with cutaneous T-cell lymphoma (CTCL) (58.0 ± 13.9 years; 26 cases of mycosis fungoides and 3 cases of Sézary syndrome), and 22 healthy control subjects (44.0 ± 17.5 years) were enrolled in the study.

Immunoreactive TSLP in sera was quantified using a human TSLP ELISA kit (R & D systems. Minneapolis, MN, USA) as previously described.[4] Statistical analysis between two groups was performed using the Mann–Whitney *U*-test.

Serum TSLP levels of patients with AD and those with psoriasis were 36.4 ± 4.2 and 35.3 ± 5.1 pg/mL, respectively (Fig. 1), and were not significantly different from healthy controls (34.2 ± 4.1 pg/mL) ($P > 0.05$). The serum TSLP level of patients with CTCL was 40.1 ± 5.0 pg/mL, which was significantly higher than that of controls ($P < 0.01$).

Serum TSLP levels greater than the mean + 2SD of the control serum samples (dotted line in Fig. 1) were considered to be raised. The percentage of increased samples was 7.7% in AD, 27.6% in CTCL and 8.6% in psoriasis.

We next studied TSLP levels at different stages of CTCL (Fig. 2). Interestingly, the percentages of increased samples were greater in the early stages (patch stage 37.5%, plaque stage 25%, tumour stage 0%, erythroderma 25%), when tumour cells have more interaction with epidermal keratinocytes. Serum TSLP levels were not correlated with age, lactate dehydrogenase levels, or other clinical and laboratory data.

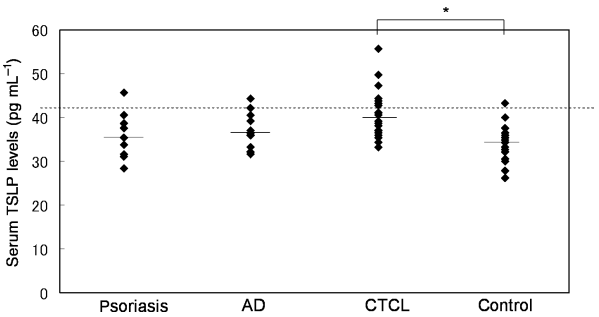

Figure 1 Serum levels of thymic stromal lymphopoietin (TSLP) in patients with psoriasis vulgaris, atopic dermatitis (AD), or cutaneous T-cell lymphoma (CTCL) and in healthy controls. Dots indicate the measured values from individual patients, with the bars being the mean of each group and the dotted horizontal line being the cut-off value (mean + 2SD of the control samples). *$P < 0.01$ by Mann–Whitney *U*-test.

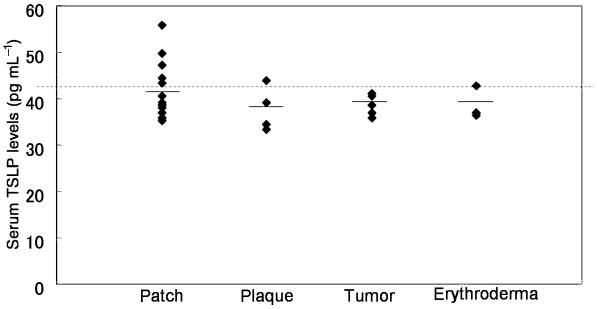

Figure 2 Serum levels of thymic stromal lymphopoietin (TSLP) in cutaneous T-cell lymphoma (CTCL) at different stages during the disease (16 cases at patch stage, 4 at plaque stage, 5 at tumour stage and four with erythroderma). Dots indicate the measured values from individual patients, with the bars being the mean of each group and the dotted horizontal line being the cut-off value (mean + 2SD of the control samples).

TSLP triggers dendritic cell-mediated Th2-type inflammatory responses and is considered to be a master switch for allergic inflammation. We found that, surprisingly, serum TSLP levels were not increased in patients with AD. In addition, the levels were not decreased in patients with psoriasis, a common Th1-type skin disease. It suggests that TSLP produced by keratinocytes may be active only in the skin, without being released in peripheral blood. Serum TSLP levels were increased in patients with CTCL, a cutaneous malignancy of lymphocytes. TSLP was recently reported to directly induce T-cell proliferation, suggesting a direct effect on lymphoma cells.[5] Because serum TSLP levels were slightly decreased as tumour cells penetrated deeper into the dermis, it is possible that tumour cells depend on TSLP from keratinocytes for their proliferation in the early stages and that they become independent at a late stage. A more detailed study would reveal the role of TSLP in the pathogenesis of CTCL.

