

A case of bullous lichen planus preceding Hodgkin's lymphoma

Maki Takada, Aki Honda, Toshiyuki Yamamoto

Department of Dermatology, Fukushima Medical University, Fukushima, Japan.

Corresponding author: Maki Takada, MD, E-mail: takadamk@fmu.ac.jp

Sir,

A 70-year-old male was referred to our department due to a progressive rash that first appeared seven months previously. He had a medical history of hypertension and hyperlipidemia, but without hepatitis B or C. A physical examination revealed scaly erythema on the upper extremities, erythema and blisters on the dorsa of the hands (Fig. 1), and hemorrhagic blisters on the lower extremities. A biopsy specimen taken from the erythema on the elbow showed irregular extensions of epidermal protrusions, lymphocytic infiltration under the epidermis, liquefaction degeneration of the basal layer, and individual cell keratinization in the epidermis (Fig. 2a). Another biopsy specimen taken from a blister on the dorsum of the hand revealed lymphocytic infiltration under the epidermis, forming subepidermal blisters (Fig. 2b). No deposition of immunoglobulins was observed in the basement membrane zone by direct immunofluorescence. Amlodipine and atorvastatin, which the patient had been receiving for hypertension and hypercholesterolemia, were discontinued, and dental metal removal was performed; however, no improvement was observed. Both topical corticosteroids and phototherapy were ineffective, and treatment with etretinate was discontinued due to liver dysfunction after only two weeks of intake. Two years after the diagnosis of bullous lichen planus (LP), he complained of general fatigue, and blood tests showed elevated C-reactive protein levels. Whole-body CT revealed multiple lymphadenopathies. A lymph node biopsy from the neck led to a diagnosis of Hodgkin's lymphoma (Fig. 3). The patient was admitted to another hospital for further treatment.

Bullous LP generally tends to appear on the oral mucosa and lower extremities, with blisters appearing



Figure 1: Clinical features of the elbow and the dorsum of the left hand.

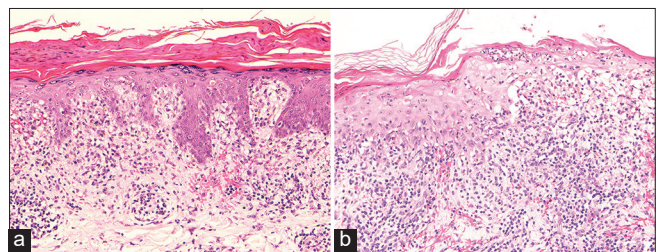


Figure 2: (a) Histopathology of the erythema on the elbow showing subepidermal lymphocytic infiltration, a poorly defined epidermal basal layer, and necrosis of epidermal cells (H&E, 200 \times). (b) Histopathology of a blister on the dorsum of the hand showing subepidermal lymphocytic infiltration and blister formation (H&E, 200 \times).

near or over existing LP lesions [1]. In the present case, blisters were seen mainly on the lower extremities. In addition, the biopsy revealed subepidermal blisters, and direct immunofluorescence revealed no deposition in the basement membrane zone. The patient was diagnosed with Hodgkin's lymphoma about two years after the diagnosis of bullous

How to cite this article: Takada M, Honda A, Yamamoto T. A case of bullous lichen planus preceding Hodgkin's lymphoma. *Our Dermatol Online*. 2026;17(2):277-278.

Submission: 14.08.2025; **Acceptance:** 13.11.2025

DOI: 10.7241/ourd.20262.31

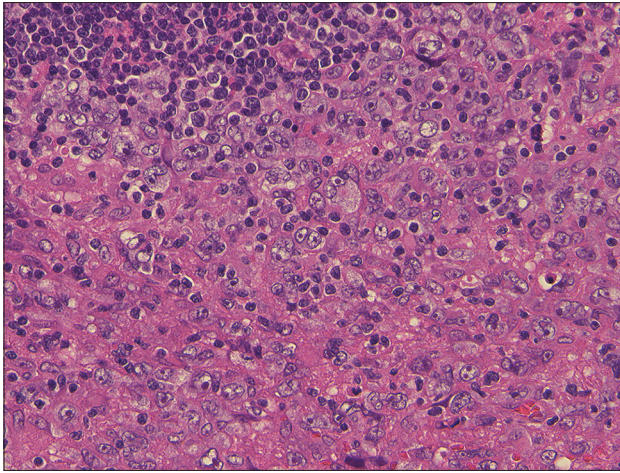


Figure 3: Atypical lymphocytes increased and Reed–Sternberg cells observed (H&E, 400x).

LP. Concurrent occurrence of LP and malignant lymphoma has been reported in only three cases in the English literature, including the present case (Table 1) [2,3]. The patients were two males and two females, and the malignant lymphomas included non-Hodgkin's lymphomas ($n = 2$) and Hodgkin's lymphoma ($n = 1$) (one was unknown). Except for the present case, malignant lymphoma preceded the occurrence of LP, and the time from the diagnosis of malignant lymphoma to LP onset ranged from 1 month to 6 years. By contrast, in the present case, LP preceded the development of the lymphoma by two years.

Various co-morbidities have been reported for LP, including hepatitis C virus infection, autoimmune diseases, internal malignancies, dyslipidemia, and viral infections [4]. Among over 13,000 reported female LP patients in Finland, 31 patients died of malignant lymphoma, among which 27 were non-Hodgkin's lymphomas and 4 Hodgkin's lymphomas [5], both showing increased standardized mortality ratios compared to the general population. Although the co-existence may be fortuitous in the present case, further accumulation of similar cases is necessary to determine the relationship between bullous LP and a hematologic malignancy.

Table 1: Report of malignant lymphoma combined with LP.

Case	Age	Sex	Types of Lymphoma	Treatment of LP	Time from Diagnosis of ML to Diagnosis of LP
1	47	F	Unknown	PSL 60 mg	6 years
2	63	F	Non-Hodgkin's lymphoma	Etretinate (1 mg/kg/day), CyA mouthwash	1 month
3	58	M	Non-Hodgkin's lymphoma	PSL 150 mg, AZP 50–100 mg, CyA 40 mg	(unknown)
Our case	70	M	Hodgkin's lymphoma	Topical steroid, oral etretinate, phototherapy	-

Abbreviations: Cya, cyclosporine; AZA, azathioprine; ML, malignant lymphoma

Consent

The examination of the patient was conducted according to the principles of the Declaration of Helsinki.

The authors certify that they have obtained all appropriate patient consent forms, in which the patients gave their consent for images and other clinical information to be included in the journal. The patients understand that their names and initials will not be published and due effort will be made to conceal their identity, but that anonymity cannot be guaranteed.

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Source of Support: This article has no funding source.

Conflict of Interest: The authors have no conflict of interest to declare.