



Case report

Burkitt lymphoma initially mimicking varicella zoster infection

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ABSTRACT

Burkitt lymphoma is an aggressive type of nodal non-Hodgkin lymphoma. This disease commonly involves the gastrointestinal tract, bone marrow, central nervous system, and in rare instances, the skin. We report the case of a 78-year-old Asian man who had experienced subacute fever and significant weight loss. He initially presented with a zosteriform skin rash on his right temporal area, which primary physicians diagnosed as varicella zoster infection and treated with antiviral drugs. The rash developed on the left forehead and chest. Dermatopathological studies on a skin lesion on the chest wall showed characteristics of Burkitt lymphoma. After a week of palliative chemotherapy, regression of all skin lesions was observed. This report aims to demonstrate an unusual zosteriform pattern of cutaneous involvement in Burkitt lymphoma.

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Introduction

Burkitt lymphoma is a highly aggressive B-cell neoplasm that commonly affects lymph nodes and extranodal sites, particularly the gastrointestinal tract, bone marrow, and central nervous system [1]. Skin involvement is rare [2–6] and exhibits short overall survival at approximately one month in patients with disseminated skin involvement [2]. A zosteriform pattern is rarely reported in patients who have developed skin metastasis with clinical zoster-like infection. We report the case of an elderly Asian man who primarily developed a skin rash with clinical presentation of dermatomal distribution on the face. The patient was finally diagnosed with skin involvement of Burkitt lymphoma.

Case presentation

A 78-year-old Asian man presented with low-grade fever for three weeks and progressively significant weight loss for six months. He noticed an asymptomatic skin rash on the right temporal area and forehead and had sought treatment at a clinic and a different hospital. Physicians diagnosed the case as varicella zoster infection and prescribed acyclovir, which did not result in clinical improvement. New skin lesions developed on the left side of the forehead and a single lesion appeared on the chest wall. The patient had no history of skin disease, including zoster infection. Physical examination revealed a group of indurated, erythematous

papules, and plaques on the right temple and forehead (distribution of trigeminal nerve, V1 dermatome) (Fig. 1A). A group of erythematous papules appeared on the left forehead (Fig. 1B) and two erythematous nodules located on the right chest wall (Fig. 1C). Bilateral cervical lymphadenopathy, approximately 1–1.5 cm in size, and a right epitrochlear node were palpated.

Skin biopsy on the chest wall was performed. The dermatopathology revealed dense infiltration of medium-sized, round nuclei-lymphoid cells with condensed chromatin in the dermis and subcutaneous tissue with a grenz zone (Fig. 2A). In some areas of the dermis, tissue macrophages engulfed apoptotic tumor cells, indicating a typical “starry sky” pattern (Fig. 2B). Immunohistochemical stainings highlighted the tumor cells with CD10, CD20, and Bcl-6, and showed negative incidence of CD3, CD5, CD56, Cyclin D1, Bcl-2, and EBER. Ki 67 proliferation index was approximately 100%. Bone marrow biopsy showed 70% cellularity and involvement of dense infiltration of medium-sized lymphoid cells with c-Myc overexpression. Computed tomography of the chest and abdomen indicated multiple enlarged nodes, approximately 0.5–2.6 cm, with circumferential thickening of the terminal ileum. Serum lactate dehydrogenase and uric acid levels were elevated at 4738 (240–480) U/L and 10.5 (3.4–7) mg/dL, respectively. Serological tests for human immunodeficiency virus, and hepatitis B and C virus were negative.

The patient was diagnosed with stage IV Burkitt lymphoma with skin involvement. He had received aggressive intravenous fluid and allopurinol to prevent tumor lysis syndrome. He had also undergone palliative chemotherapy (cyclophosphamide, doxorubicin, vincristine, methotrexate, and prednisolone) based on advanced age and poor performance status. All skin lesions

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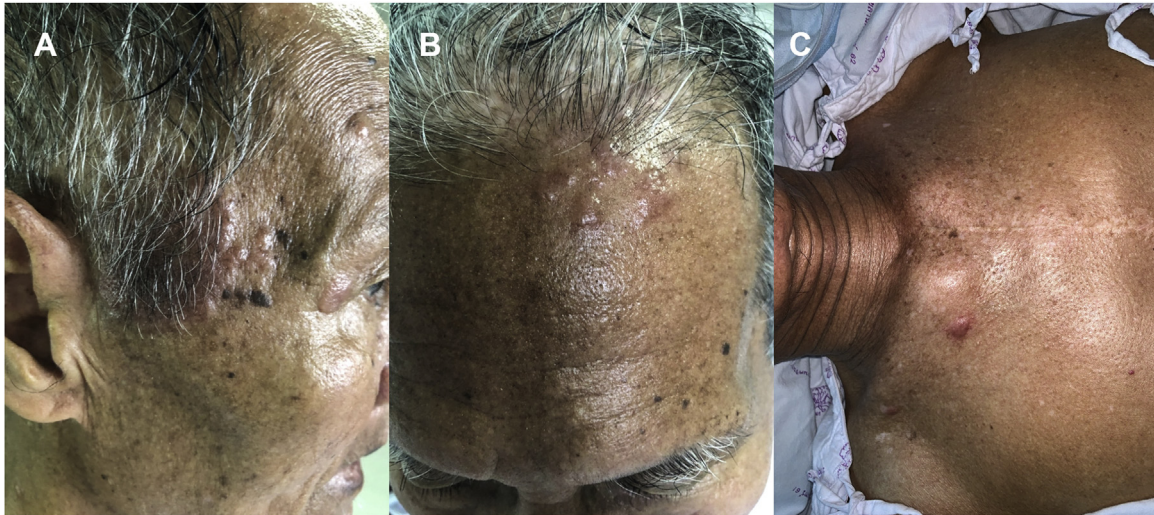


Fig. 1. Clinical features of Burkitt lymphoma skin lesions: A) group of erythematous papules and plaque on right temple and forehead (trigeminal nerve, V1 dermatome), B) group of erythematous papules on left forehead, and C) two erythematous nodules on right chest wall.

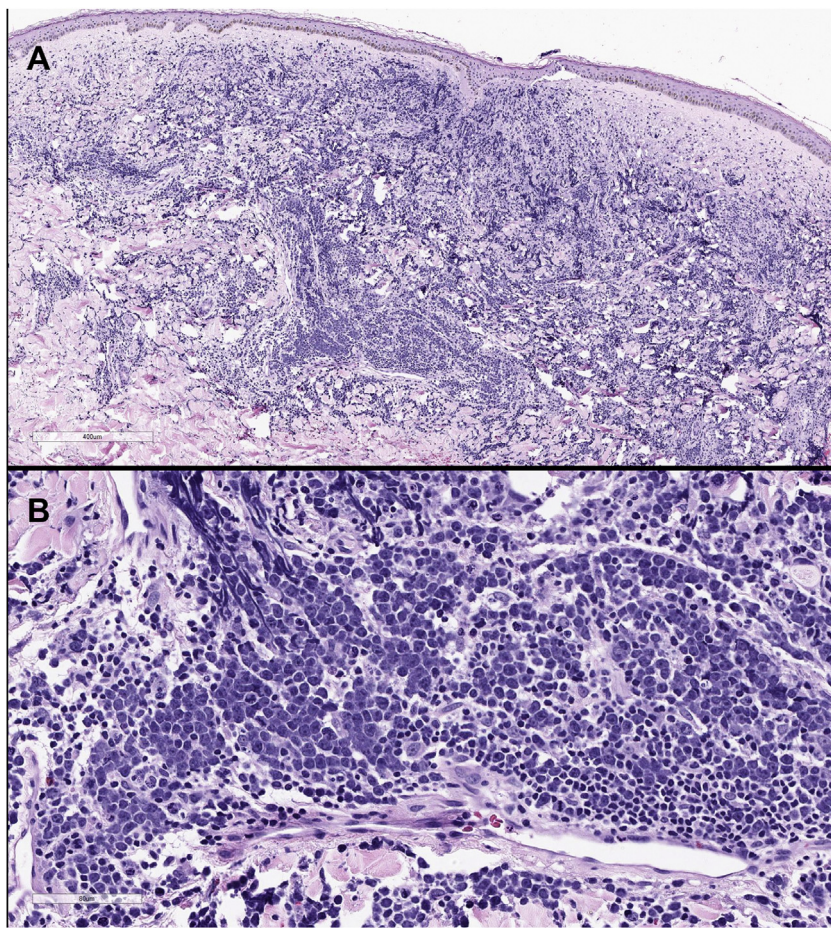


Fig. 2. Microscopic examination of skin biopsy specimen on chest (hematoxylin and eosin staining): A) low power revealed dense infiltration of medium-sized, round nuclei-lymphoid cells with condensed chromatin in the dermis and subcutaneous tissue with a grenz zone ($\times 60$); and B) tissue macrophages engulfed apoptotic tumor cells, which indicate a typical "starry sky" pattern ($\times 300$) in some areas of the dermis.

disappeared after a week of treatment. However, the patient developed tumor lysis syndrome and febrile neutropenia. Four months after the diagnosis, the patient died due to the progression of the disease.

Discussion

Burkitt lymphoma has three main clinical variants: endemic, sporadic, and immunodeficiency-related. The endemic form, which originated from Africa, is associated with Epstein-Barr virus and commonly manifests in the jaw, ovaries, breasts, and kidneys [1]. The sporadic and immunodeficiency-related variant frequently affects the gastrointestinal tract, bone marrow, and central nervous system [1].

Cutaneous involvement of Burkitt lymphoma is extremely rare [2–6]. Previous case reports have revealed the route of skin metastasis in various ways, such as hematogenous spreading, local invasion from the lymph nodes, trauma, or surgical procedure [3–6]. The zosteriform pattern is rarely reported in some primary malignancies, for example, lung cancer, eccrine porocarcinoma, melanoma, colon cancer, and hematologic malignancy, including T-cell and B-cell lymphomas [7–12]. The proposed pathogenesis was related to previous traumatic skin disorders, including zoster infection. To the best of our knowledge, this is the first case report on Burkitt lymphoma that initially presents with zosteriform metastasis. However, the patient's skin lesions progressed to other areas. The cutaneous metastasis should be through the hematogenous route.

In the treatment context, the standard regimens for Burkitt lymphoma are CODOX-M/IVAC (cyclophosphamide, doxorubicin, vincristine, methotrexate/ifosfamide, cytarabine, and etoposide), DA-EPOCH-R (dose-adjusted, etoposide, prednisone, vincristine, cyclophosphamide, doxorubicin, and rituximab), and hyper-CVAD (hyper-hyperfractionated cyclophosphamide, vincristine, doxorubicin, and dexamethasone), which can achieve complete response [1]. These regimens are not well tolerated by older adults with poor performance status, and the patients experience toxicity from the standard treatment.

In conclusion, cutaneous involvement of Burkitt lymphoma is infrequent. The zosteriform metastasis is uncommon in primary solid and hematologic neoplasm. This study reported the first case of Burkitt lymphoma with cutaneous involvement that initially mimicked zoster infection.

CRediT authorship contribution statement

Nuntouchaporn Amonchaisakda: Conceptualization, Data curation, Writing - original draft. **Kumpol Aiempanakit:** Conceptualization, Data curation, Supervision, Writing - original draft, Writing - review & editing. **Benjawan Apinatriyo:** Supervision, Writing - review & editing.

Declaration of Competing Interest

There are no conflicts of interest.

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