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CASE REPORT

Hypersensitivity to mosquito bites as the primary clinical manifestation of an Epstein–Barr virus infection



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Hypersensitivity to mosquito bites (HMB) is a rare disease characterized by intense local skin reactions with general symptoms, such as high fever and regional lymphadenopathy after mosquito bites. Epstein–Barr virus (EBV) chronic infection and NK cell lymphoproliferative disease have been reported first in diagnosed HMB patients. Here, we present the case of a 6-year-old girl with 2 months' history of bullae and necrotic skin lesions, accompanied by a high temperature, visual hallucinations, and liver dysfunction after mosquito bites. A histopathologic examination of the skin lesion showed vasculitis and EBV infection. We could not detect any findings of hematologic malignancies or NK cell proliferative disease in the patient. Clinicians should closely evaluate HMB patients for possible development of lymphoproliferative status or hematologic malignant disorders.

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Introduction

Hypersensitivity to mosquito bites (HMB) is characterized by intense local skin reactions with general symptoms, such as high fever and regional lymphadenopathy after mosquito bites.¹ Most patients present with clinical symptoms in the first two decades of life, with a median age of 6.7 years. This disorder has a strong racial predisposition and few case

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reports of HMB have been reported from Japan and Korea.^{2,3}

HMB patients have been reported with chronic Epstein–Barr (EB) virus infection and natural killer (NK) cell leukemia/lymphoma. The pathogenesis of HMB may be related to clonal lymphoproliferation of EB virus DNA-positive NK cells.² Here, we describe a patient with systemic EBV infection who presented with hypersensitivity to mosquito bites but without NK cell proliferative disease.

Case report

A 6-year-old girl was referred to our hospital with three episodes of systemic symptoms including a high temperature rising to 40°C, and visual hallucinations after mosquito bites. Physical examination revealed bullae and subsequently necrotic skin lesions at the mosquito bite sites (Fig. 1). Hepatosplenomegaly and peripheral lymphadenopathy was not detected. She had a normal life during symptom-free days and did not have a family history of similar conditions. Abnormal EEG recording with epileptogenic activity over the left occipital area was noted. No definite abnormality was found by chest X-ray and brain magnetic resonance imaging. Laboratory tests showed the following results: white blood cell 11,100/mL (neutrophil 61%, lymphocyte 23%), C-reactive protein (2.55 mg/dL), erythrocyte sedimentation rate (10 mm/hour), C3 (140 mg/dL) and C4 (34.5 mg/dL). High IgE level with 1784 kIU/L and liver function impairment with ALT (680 U/L), AST (136 U/L) were found. IgG (1160 mg/dL), IgA (121 mg/dL), IgM (57.4 mg/dL) levels, and lactate dehydrogenase were normal. CSF examination did not show positive findings. Anti-neutrophil cytoplasmic antibody and anti-nuclear antibody were negative. Lymphocyte subset analysis demonstrated percentages of CD3 (78%), CD4 (43.1%), CD8 (31.6%), NK cell (3.2%), and CD19 (14%). IgM for anti-nuclear antigen (EBNA), viral capsid antigen (VCA) and anti-early antigen (EA) to EBV were all negative. However, the levels of anti-VCA IgG, anti-EA-DR IgG and anti-EBNA IgG were increased which is consistent with chronic EBV infection. A skin biopsy from the lesion showed necrosis, interstitial and perivascular eosinophilic and lymphocytic infiltrate, and small vessels with fibrinoid necrosis (Fig. 2A). *In situ* hybridization for EBER (EBV encoded RNA) was positive (Fig. 2B) and NK cell marker (CD56) was negative

(Fig. 2C) in the mosquito bite site. The patient had a bone marrow biopsy, and EBER revealed a positive finding (Fig. 2D). The skin lesion and systemic symptoms improved with treatment with oral corticosteroids. We could not detect any findings of hematologic malignancies.

Discussion

Hypersensitivity to mosquito bites (HMB) was initially thought to be a simple allergic reaction to mosquito bites; however, it showed several features distinct from common mosquito bite allergies. In patients with HMB, the cutaneous reaction such as bullae and necrotic ulcerations at bite sites is much more severe compared with the general skin lesion induced by mosquito bites and is further associated with systemic symptoms such as high-grade fever and peripheral lymphadenopathy.^{1–3} In our case, the clinical features with HMB included periodic attacks with intense local skin reactions accompanied by high fever, high IgE level, and perivascular eosinophilic and lymphocytic infiltrate with fibrinoid necrosis in mosquito bites sites, compatible with hypersensitivity vasculitis.

Epstein–Barr virus (EBV) is a member of the herpesvirus family and has occasionally been implicated in the pathogenesis of leukocytoclastic and granulomatous vasculitis, lymphocytic vasculitis, and granulomatous vasculitis.^{4,5} As cases accumulated, it became apparent that up to 33% of HMB patients have been associated with chronic active EBV infection (CAEBV).² The case did not present clinical symptoms and blood smear regarding EBV associated infectious mononucleosis and no hemophagocytic lymphohistiocytosis (HLH) was noted by bone marrow aspiration. We present this case with visual hallucinations and distortion of the body image with characteristics of “Alice in Wonderland” syndrome by EBV infection. Electroencephalography showed focal spikes over the left occipital areas and may be the cause of the visual hallucinations. Visual hallucinations are a peculiar characteristic of primary EBV infection and may be due to recently clinical presentation accompanied by HMB. In this case, the patient had visual hallucinations, liver dysfunction, elevated EBV serum titer, and positive EBER findings *in situ* hybridization in mosquito bite sites and bone marrow biopsy which is consistent with chronic EBV infection.

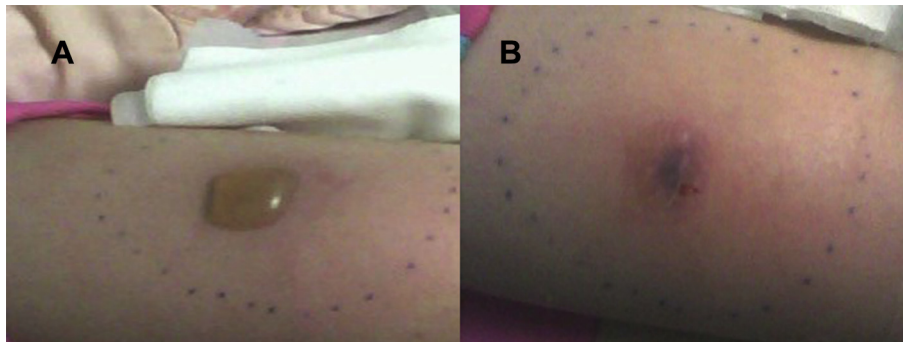


Figure 1. Skin lesion at a mosquito bite site on the left forearm showing (A) erythema, swelling and bullae and (B) subsequent necrosis formation.

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