A case of lymphomatoid granulomatosis presenting with cutaneous lesions

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INTRODUCTION

Lymphomatoid granulomatosis (LYG) is a rare B-cell lymphoproliferative disorder that involves the skin in approximately 50% of cases. We describe a patient with LYG who first presented with cutaneous lesions. His skin biopsy failed to show large B-cell lymphocytes positive for Epstein-Barr virus (EBV)-encoded RNA in situ hybridization (EBER-ISH), highlighting the diagnostic challenges that dermatologists may face when encountering cutaneous LYG.

Abbreviations used:

CT:	computed tomography
EBER-ISH:	Epstein-Barr virus—encoded RNA in situ hybridization
EBV:	Epstein-Barr virus
LYG:	lymphomatoid granulomatosis

CASE REPORT

A previously healthy 28-year-old man presented with recurrent nontender, erythematous skin lesions lasting several months, associated with intermittent fever, weight loss, and cough of 1-month duration. Prior workup included a punch biopsy by an outside dermatologist interpreted as granulomatous dermatitis, chest computed tomography (CT) showing bilateral noncalcified pulmonary nodules, a nondiagnostic CT-guided lung biopsy, and a normal bone marrow biopsy.

On examination, he had multiple nontender, subcutaneous nodules and nummular erythema (Fig 1) scattered throughout the trunk and upper and lower extremities. Skin biopsy findings showed an atypical dense dermal lymphohistiocytic infiltrate predominantly surrounding the blood vessels (Fig 2). Immunohistochemical staining found an infiltrate of predominantly $CD4^+$ lymphocytes; some small $CD20^+$ B cells surrounding blood vessels; and rare $CD30^+$, $CD15^-$ large lymphocytes.

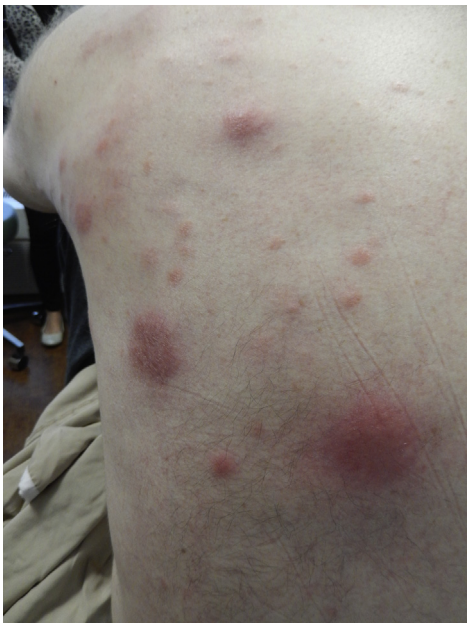

Fig 1. Lymphomatoid granulomatosis as seen on a previously healthy 28-year-old man. Multiple yellowish-red, 3-mm to 4-cm papules and circular plaques over the left back.

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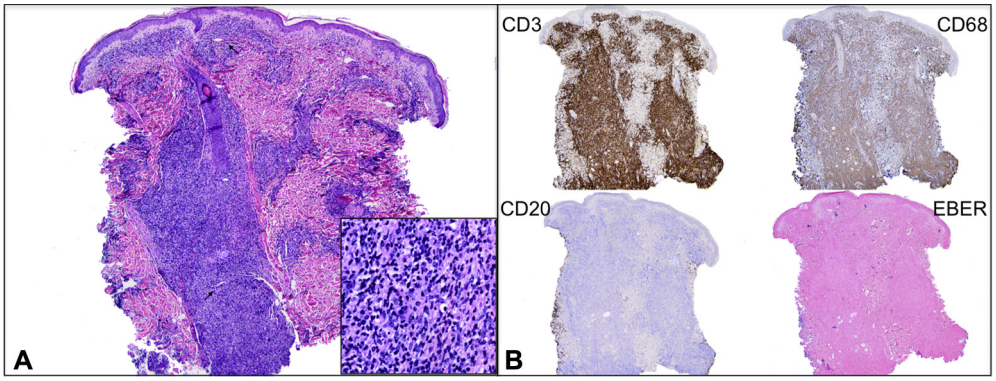

Fig 2. Lymphomatoid granulomatosis as seen on a skin biopsy specimen from the right upper arm. **A**, A dense dermal nodular lymphoid infiltrate surrounding the vessel walls (*black arrows*). Inset shows an inflammatory infiltrate with pleomorphic morphology and consisting of atypical-appearing lymphocytes associated with a few large/transformed cells and a prominent histiocytic component in the background. **B**, Immunohistochemical analysis of skin biopsy shows an infiltrate that is diffusely and strongly positive for CD3, diffusely positive for CD68, very focally and weakly positive for CD20 and negative for EBER-ISH. (**A** and **inset**, Hematoxylin-eosin stain; original magnifications: A, ×4; inset, ×40; **B**, Immunohistochemical staining for CD3, CD68, CD20 antibodies and in-situ hybridization for Epstein-Barr virus-encoded small RNA; original magnification: ×4.)

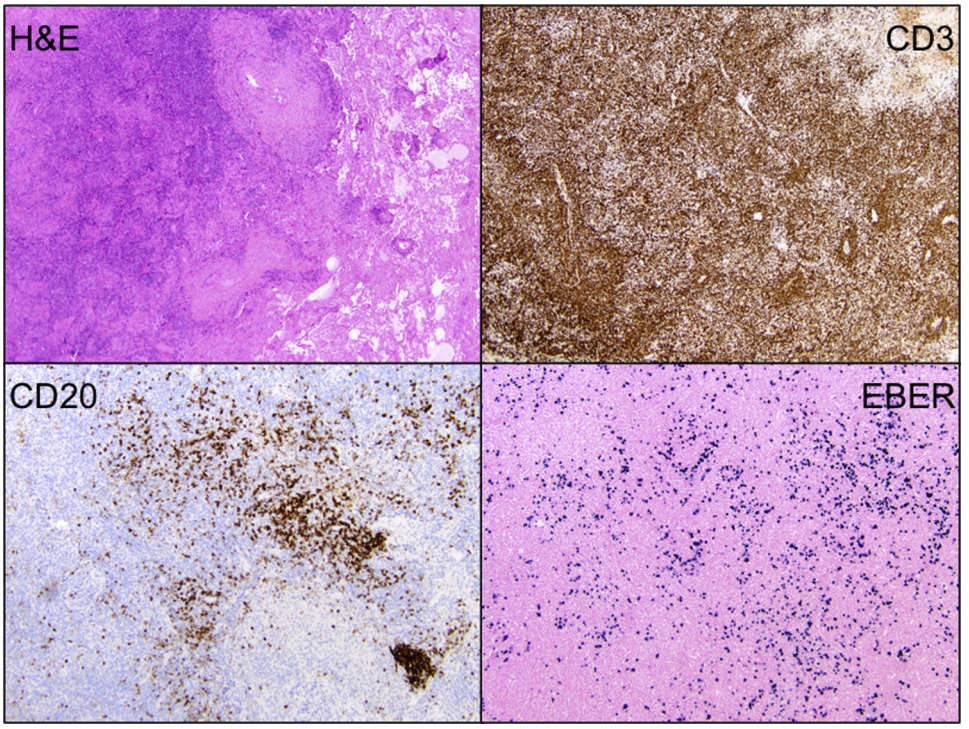

Fig 3. Lymphomatoid granulomatosis as seen on H&E staining and immunohistochemical analysis of lung biopsy. The infiltrate is strongly positive for CD3 and CD20. EBER-ISH is positive in atypical lymphocytes present in lung tissue (EBER positivity is indicated by dark blue staining of the nuclei). (Hematoxylin-eosin stain; original magnification: ×4.)

$CD20^+$ cells were a minor component and results of EBER-ISH were negative. T-cell receptor beta gene rearrangement by florescent polymerase chain reaction showed a polyclonal pattern, militating against a T-cell lymphoproliferative disorder.

Several months later, the patient underwent a lung wedge resection because of worsening constitutional symptoms. Pathologic evaluation of the lung specimen found an atypical lymphohistiocytic infiltrate, with $CD20^+$ lymphocytes, which were