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SHORT REPORT

EBV-associated lymphoproliferative disorders misdiagnosed as Crohn's disease

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Abstract

Epstein–Barr virus (EBV) plays an etiological role in various diseases. EBV-associated lymphoproliferative disorder (LPD) is usually observed in individuals with congenital or acquired immune deficiencies but was also recently reported in non-immunocompromised individuals. Two cases of immunocompetent patients with EBV-associated T-cell LPD of the small bowel and colon who were initially misdiagnosed as Crohn's disease (CD) are reported here. EBV-associated T-cell LPD with primary gastrointestinal tract involvement can manifest as multiple discrete ulcers of the small and/or large bowel that are similar to the lesions found in CD or intestinal tuberculosis. However, when patients have multiple intestinal ulcers that are not typical of CD or intestinal tuberculosis and the clinical course is unusual, clinicians should consider the

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possibility of EBV-associated LPD that involves the gastrointestinal tract because the treatment strategy and prognosis are completely different.

1. Introduction

Epstein–Barr virus (EBV) plays an etiological role in various diseases, including infectious mononucleosis, chronic active EBV infection, malignancies such as nasopharyngeal cancer and Burkitt's lymphoma.[1,2] EBV-associated lymphoproliferative disorder (LPD) is usually observed in individuals with congenital or acquired immune deficiencies, particularly patients who have undergone solid organ or hematopoietic transplantation.[3,4] Recently, however, EBV-associated LPD has been reported in non-immunocompromised individuals.[5] It is very rare to encounter immunocompetent patients with EBV-associated LPD involving gastrointestinal tract primarily. The cases of two immunocompetent patients with EBV-associated T-cell LPD of the small bowel and colon who were initially misdiagnosed as Crohn's disease (CD) are described here.

2. Case report

2.1. Case 1

A previously healthy 50-year-old man with no personal or family history of immunodeficiency presented with an 8-year history of loose stools (1–3 times per day) and intermittent febrile sensation. He had been diagnosed with intestinal tuberculosis (TB) 5 years earlier when multiple ulcerations in the colon were detected. When standard first-line anti-tuberculous medication was administered for 9 months, he showed some clinical improvement. Four years ago, he visited the department of oncology at our hospital because of recurrent symptoms. Various examinations, including colonoscopy, abdominal computed tomography (CT) and positron emission tomography (PET)-CT, were performed. Multiple active ulcers were observed on colonoscopy but a definite diagnosis could not be made. Although it was recommended that he should be evaluated further to rule out the possibility of colonic LPD, he refused and was lost to follow-up. After that, at another hospital, he was treated with second and third courses of first-line anti-tuberculous medication for 9 and 18 months, respectively. Since colonic ulcers persisted during the third course, the diagnosis was revised to CD. Prednisolone and mesalamine were prescribed but he did not take these medications and visited our hospital for a second opinion.

At presentation, he complained of general weakness, anorexia, weight loss (12 kg over 2 months), loose stools, and fever. The physical examination revealed mild tenderness on both lower abdomen quadrants. Laboratory tests showed mild leukocytosis (11,600/mm^3), anemia (11.8 g/dL), increased C-reactive protein (CRP) levels (4.46 mg/dL), hypoalbuminemia (1.8 g/dL), and elevations of the liver enzymes aspartate aminotransferase (58 IU/L) and alanine aminotransferase (56 IU/L). The stool assays were all negative.

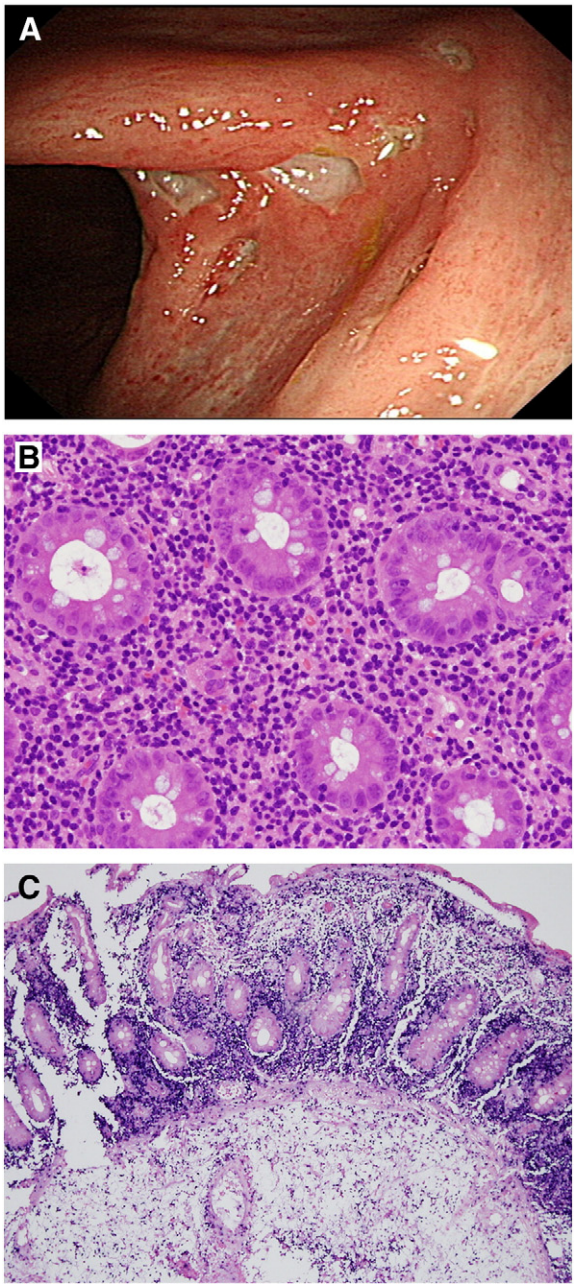

Figure 1 Endoscopic and histologic findings of case 1. (A) Colonoscopic finding. It shows multiple well-demarcated deep ulcers with clean bases. (B) Histopathology of a colon biopsy. It shows infiltration of atypical, hyperchromatic lymphoid cells (H&E stain, ×200). (C) EBV in situ hybridization. It shows marked infiltration of EBV-positive cells into the mucosa and submucosa (×40).