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An Atypical Extraintestinal Manifestation in a Child with Ulcerative Colitis: Cutaneous Leukocytoclastic Vasculitis

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Dear Editor:

Approximately 15%～20% of the pediatric patients with inflammatory bowel disease develop extraintestinal manifestations[1,2]. The most common cutaneous extraintestinal manifestation of ulcerative colitis (UC) is erythema nodosum, pyoderma gangrenosum, and psoriasis[2]. Leukocytoclastic vasculitis (LCV) can rarely be associated with UC and it is reported that the onset of cutaneous LCV precedes the intestinal symptoms[1]. Here, we present a case diagnosed with LCV 2 weeks after the development of pediatric UC.

A 14-year-old girl was admitted to our hospital for evaluation of sudden pain of left shoulder and intestinal symptoms such as abdominal pain, nausea, vomiting and diarrhea for 2 days. Two days later, she presented with bloody diarrhea 2～3 times a day. Colonoscopy revealed edematous mucosal erythema and exudates, mucosal friability, and spontaneous bleeding on the entire colon and the rectum. Magnetic resonance imaging of left shoulder showed possible multifocal myositis of left shoulder. Laboratory findings were within normal limits, except for elevated white blood cell count (10,700/ml) and C-reactive protein levels (7.92 mg/dl). Serologic examination was positive for perinuclear antineutrophil cytoplasmic antibodies. Methylprednisone was initiated at a dose of 1 mg/kg/d, resulting in rapid relief from myalgia and hematochezia. Two weeks later, she was referred to the department of dermatology for tender, purpuric patches and bullae on her left buttock, right shoulder and lower abdomen (Fig. 1). A biopsy specimen of right shoulder demonstrated LCV (Fig. 2). Cyclosporine was started intravenously at 2 mg/kg/d; subsequently, the skin eruption and intestinal symptoms slowly resolved. She was still well with oral mesalazine and azathioprine.

LCV is characterized by inflammation of postcapillary venules with neutrophilic infiltration, nuclear debris and fibrinoid necrosis. It is considered as an immune-complex disorder triggered by various drugs, infections, malignancies, and autoimmune disorders[1]. LCV is uncommon in patients with UC as compared to other skin manifestations[2]. One possible explanation of the association between these two disorders is that the pathogenesis of both is based on immune mechanisms and deposition of immune complexes in the vascular structure and intestinal mucosa for LCV and UC, respectively[3]. In the English literature, 19 cases of biopsy-proven LCV have been reported in patients with UC[4,5]. In 8 out of 20 patients (40%) with UC, LCV developed before 20 years of age. In 7 out of 20 patients (35%), LCV developed prior to clinical manifestations of UC by 1-18 months. LCV and UC occurred concurrently in 4 patients (20%). Nine out of 20 patients (45%) had a diagnosis of UC before presenting LCV from 2 weeks to 20 years. Lower extremities were the most commonly involved sites of LCV (90%). In most cases, cutaneous symptoms presented as purpuric macules or patches. As the treatment for these two disorders involves same medications, LCV complicating UC can be treated with corticosteroids or immunosuppresive agents.

In conclusion, dermatologists should be aware that LCV may rarely occur before, after, or simultaneously with the diagnosis of UC, as it does not show a typical symptom

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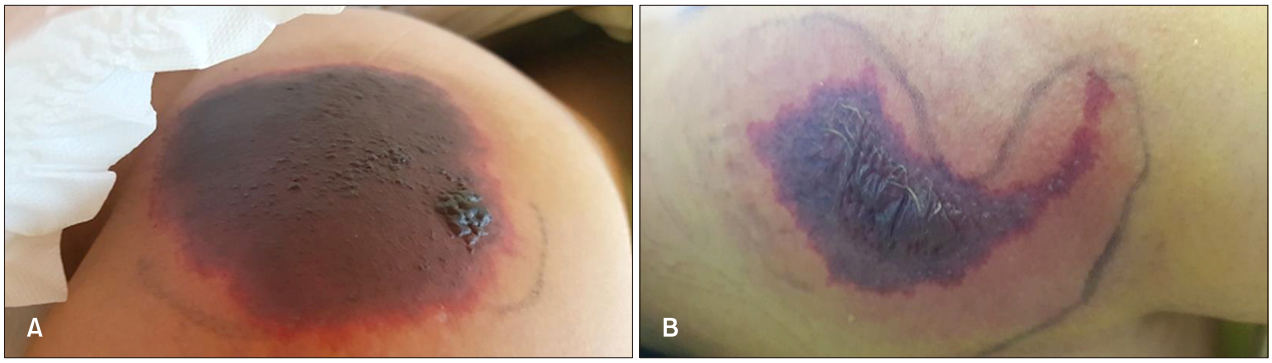

Fig. 1. (A, B) Physical examination showed tender, purpuric patches and bullae on her left buttock (A) and right shoulder (B). We received the patient's consent form about publishing all photographic materials.

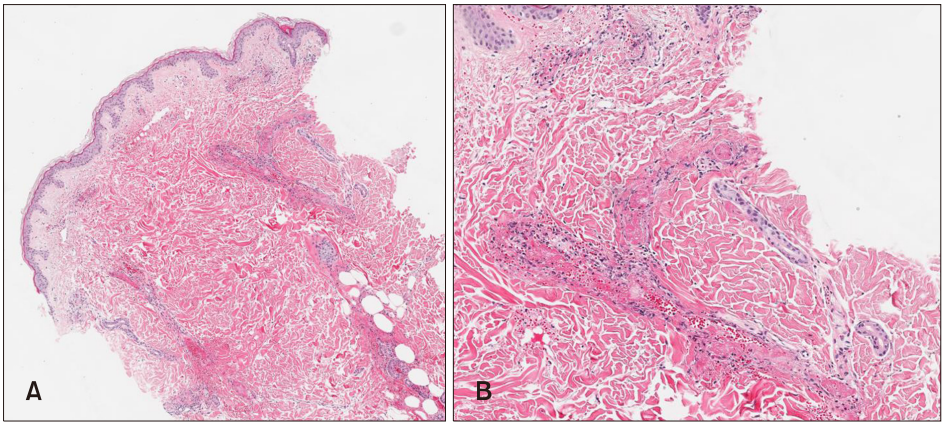

Fig. 2. (A, B) Histopathological findings revealed leukocytoclastic vasculitis. Fibrinoid necrosis of vessel wall, neutrophilic infiltration and extravasation of red blood cells were shown (A: hematoxylin and eosin [H&E], ×40; B: H&E, ×200).

and leaves a permanent defect if early treatment is not presented.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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