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Granulomatous Variant of Food Protein-Induced Allergic Proctocolitis

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A 7-month-old infant on commercial cow milk formula presented with a 4-week history of bloody stools without vomiting, diarrhea, fever, or skin rash. The patient did not appear ill, had a normal physical examination, and took his feeds well. Laboratory results showed a leukocyte count of 17,400/mm^3 (reference range, 4400–19,100/mm^3) with 12% eosinophils (0.0%–5.0%), hemoglobin level of 12.7 g/dL (10.1–14.2 g/dL), and albumin level of 4.3 g/dL (3.2–4.8. g/dL). Stool cultures were unremarkable. A stool smear showed a large number of eosinophils. Serum IgE, IgG, IgM, and IgA levels were unremarkable, and radioallergosorbent test, performed to determine the presence of specific IgE against cow milk, α-lactalbumin, or casein were all negative.

Rectosigmoidoscopy revealed patchy erythema with congestion of the rectal and sigmoid colon mucosa (Fig. 1). Biopsy specimens revealed numerous eosinophils and degranulation in the mucosa (Fig. 2), and notably, granulomatous infiltrates composed of multinucleate giant cells with prominent eosinophils and degranulation in the submucosa (Fig. 3).

Immunodeficiency was considered, but history of unusual, severe, or recurrent infections was negative, and serum immunoglobulin levels were normal. Although presentation with granulomas is atypical, the patient was diagnosed with food protein-induced allergic proctocolitis (FPIAP) based on the clinical and histological findings. The commercial cow milk formula was replaced with an extensively hydrolyzed formula, resulting in a dramatic improvement in the condition of the patient. The patient has remained on an extensively hydrolyzed formula and has been consuming a variety of solid foods. No relapse of symptoms has been observed, 5 months after the diagnosis.

FPIAP is a common cause of rectal bleeding in an otherwise healthy young infant (1). In most cases of FPIAP, biopsy specimens show prominent eosinophilic infiltration of the rectosigmoid mucosa (2,3). Occurrence of a granulomatous component, as seen in this

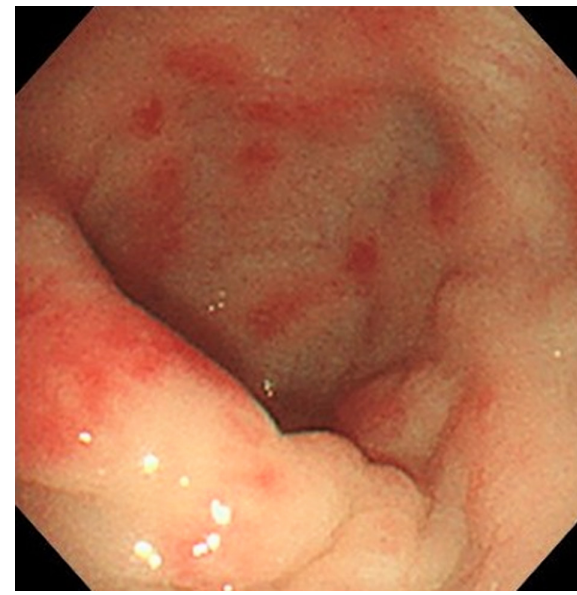

FIGURE 1. Rectosigmoidoscopy results showing patchy erythema with congestion of the mucosa in the rectum and sigmoid colon.

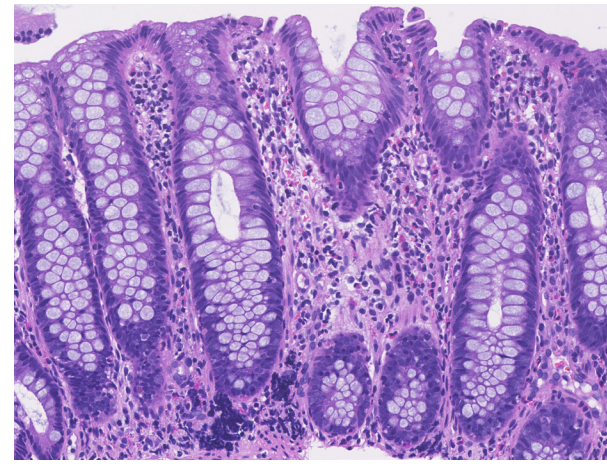

FIGURE 2. Biopsy specimens showing numerous eosinophils and degranulation in the mucosa of the sigmoid colon (hematoxylin and eosin staining; original magnification, 200X).

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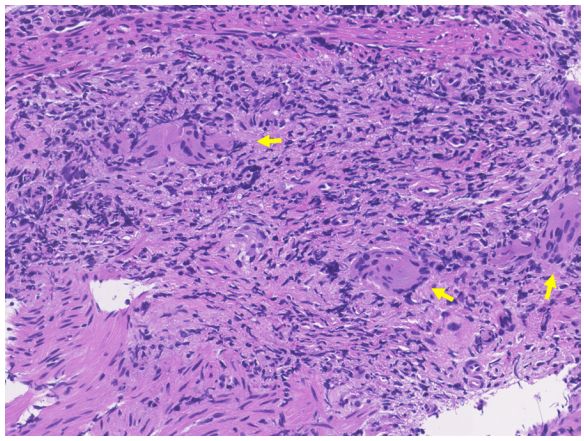

FIGURE 3. Biopsy specimens showing granulomatous infiltrates composed of multinucleate giant cells with prominent eosinophils and degranulation in the submucosa of the sigmoid colon (arrow: multinucleate giant cell, hematoxylin and eosin staining; original magnification, 200X).

patient, is a distinctly rare phenomenon in this disease. To date, only two studies have shown the presence of granulomas in FPIAP (4,5). The presence of a granulomatous infiltrate may indicate a severe form of FPIAP (4). Generally, the prognosis of FPIAP is favorable, and tolerance is acquired by around the 1-year of age (6,7). The presence of granulomas in FPIAP should not lead to a diagnosis of any other disease that presents with granulomas based on histological findings, such as early-onset inflammatory bowel disease, primary immunodeficiency, or various infectious diseases (8). Awareness of such unusual and misleading features in FPIAP is, therefore, necessary to avoid unfortunate misdiagnosis as another disease.

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