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CASE REPORT

Eosinophilic esophagitis, gastroenteritis, and colitis in a patient with prior parasite exposure

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Abstract
Eosinophilic gastrointestinal disorders (EGID) comprise a spectrum of inflammatory diseases that can affect any segment of the gastrointestinal tract. The pathogenesis of these conditions is complex; differentiating between primary and secondary forms of these disorders can be clinically challenging. We report a case of primary EGID in a patient with remote parasite exposure, whose symptoms were initially attributed to irritable bowel syndrome. Endoscopy revealed the rare finding of EGID involving the entire gastrointestinal tract; symptoms improved with an elimination diet. This case raises the possibility of a link between prior parasite exposure and development of EGID, and underscores the necessity of exploring alternative diagnoses in patients with presumed IBS who present with severe symptoms.

Keywords Eosinophilic · Esophagitis · Gastroenteritis · Colitis

Introduction

Eosinophilic gastrointestinal disorders (EGID) represent a spectrum of inflammatory diseases characterized by eosinophil infiltration throughout the gastrointestinal (GI) tract [1–3]. The prevalence of EGID varies, with eosinophilic esophagitis (EoE) being the most common (0.4% of the Western population) [4]. Eosinophilic gastroenteritis (EGE), however, is much less common with an estimated prevalence of 0.008%; eosinophilic colitis (EC) is even rarer (0.003% in the United States) [5–7]. Pathogenesis and risk factors for EGID are poorly understood, and diagnosis remains challenging. While formal diagnostic criteria exist for EoE, they are lacking for EGE and EC [4, 8–10]. We report a case of primary EGID in a patient with prior parasite exposure.

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Case report

A 58-year-old Caucasian man, previously diagnosed with irritable bowel syndrome, was admitted with increasing diarrhea, emesis, and 30-pound weight loss. Two years prior to admission, esophagogastroduodenoscopy (EGD) and colonoscopy were notable for gastroduodenitis. Three months prior, he began experiencing approximately 10 watery bowel movements per day, along with emesis and cramping abdominal pain. These symptoms progressively worsened, resulting in syncope and acute kidney injury due to volume depletion, and he was admitted to the hospital. He denied fevers, dysphagia, hematochezia, rash, or new medications.

On the floor, physical examination disclosed normal vital signs and abdominal tenderness in the lower quadrants. Notable initial studies included a white blood cell count of 13,400/µl (15% eosinophils; absolute eosinophil count 2050/µl), erythrocyte sedimentation rate 33 mm/h, C-reactive protein 22 mg/dl, and normal lipase. Stool studies revealed positive occult blood, low fecal elastase (32 µg/g; normal > 200 µg/g) and elevated fecal calprotectin (246 µg/g; normal < 163 µg/g). Stool culture, ova and parasites, and *C. difficile* toxin PCR were negative. Toxocara antibody titer was 1:8, which was below the positive cutoff as specified by the Center for Disease Control (< 1:32). CT scan of the abdomen revealed hepatic steatosis, gallbladder sludge, and pancreatic calcifications.

Further review of the electronic medical record revealed peripheral eosinophilia since 2015, with peak absolute eosinophil count 3630/µl. Additional results from the present admission included elevated serum IgE (> 10,000 U/ml) and normal flow cytometry. The patient tested negative for HIV and HTLV-1, as well as for mutations in JAK2, BCR/ABL, and FIP1L1-PDGFRA.

Empiric trial of pancreatic enzyme supplementation was unsuccessful. Subsequent EGD revealed esophagitis, gastritis, duodenitis (Fig. 1); colonoscopy revealed normal-appearing mucosa. Random biopsies from the upper and lower GI tract revealed marked eosinophilic infiltrate in all samples without evidence of parasitic infection (Fig. 2). The gastroenterology consultants recommended transition to a lactose-free, low-residue diet. The infectious diseases consultants considered 4 weeks of albendazole followed by corticosteroid therapy; however, prior to receiving any treatment, the patient's symptoms improved with dietary modification. According to the patient, dairy avoidance was most effective for his symptoms. Five months later, the serum absolute eosinophil count improved to 690/µl, and further decreased to 430/µl by 10 months.

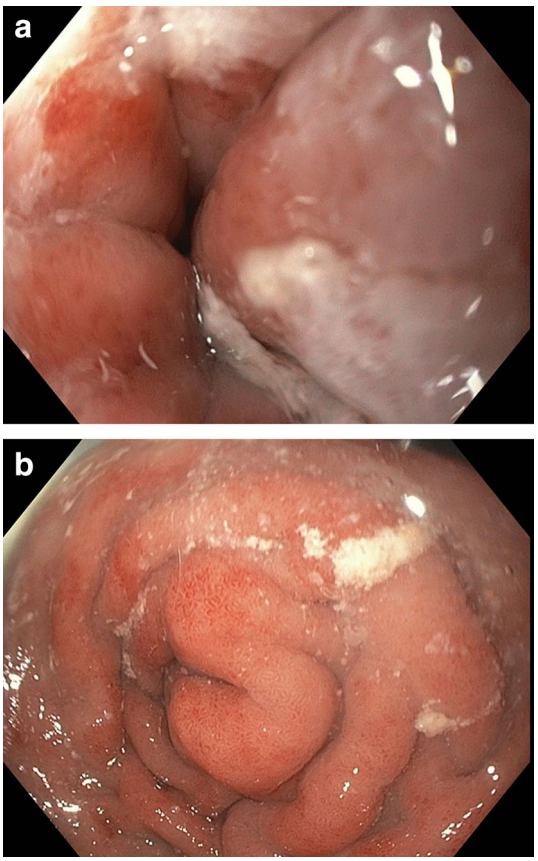

Fig. 1 Endoscopic views from EGD. **a** Gastroesophageal (GE) junction: diffuse LA grade C esophagitis. **b** Duodenum: congested mucosa and erythema consistent with duodenitis

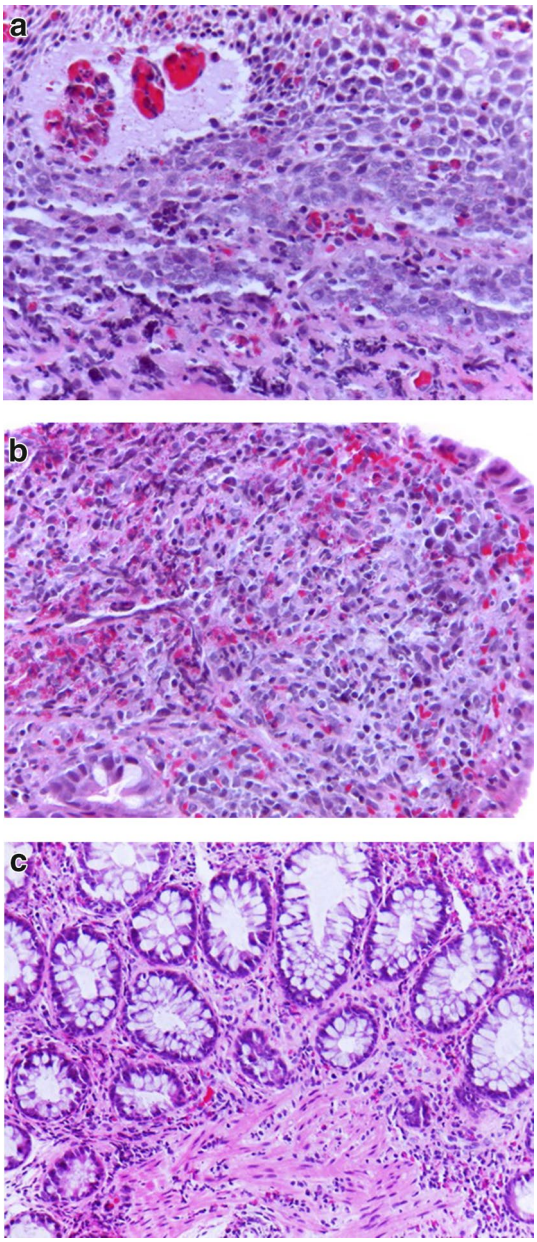

Fig. 2 Histopathology of biopsy samples from EGD and colonoscopy. **a** GE junction: eosinophilic infiltration with spongiosis and microabscess formation. **b** Duodenum: villous blunting with massive eosinophilic infiltrate, degranulation, and sheets of lymphoplasmacytes. **c** Left colon: dense eosinophilic infiltrate in the full thickness of lamina propria, focally involving the muscularis mucosae. Eosinophil count in each image is > 100 per high-power field

Discussion

Primary EGID is rising in prevalence and incidence [11, 12]. Approximately 200 case reports have described EC, and fewer than 10 have discussed EoE, EGE, and EC occurring simultaneously [4, 5, 13]. The pathophysiology

is poorly understood and is multifactorial; genetics, atopy (e.g. food allergy), and adaptive T cell immunity are thought to contribute [2, 14, 15]. At diagnosis, primary EGID must be differentiated from secondary causes such as parasitic infection, hypereosinophilic syndrome [16], vasculitis, drug reaction, and malignancy [14, 15]. Of note, peripheral eosinophilia may not be present and is not required for the diagnosis of EGID [14, 15]. Radiographic findings are variable and nonspecific; CT may be normal or may show bowel wall thickening, dilation, or prominent mucosal folds [17]. Histopathology samples of normal GI mucosa show an upper limit of normal of 6–20 eosinophils per high-power field (HPF) [18, 19]; patients with EGID often demonstrate marked elevations in the rage of 40–1200 eosinophils per HPF [19].

Initial management of primary EGID consists of an empiric trial of an elimination diet [2, 5, 6]. Systemic corticosteroid therapy may be required in severe cases or if conservative measures have failed [2, 5]. Adjunctive agents such as montelukast (leukotriene receptor antagonist), ketotifen (anti-histamine), and cromolyn (mast cell stabilizer) may be beneficial [2, 6]. More recently, attention has shifted to biologics, including monoclonal antibodies targeting IL-5 such as reslizumab and mepolizumab. Several trials indicate that these agents may be helpful in corticosteroid-refractory disease [2, 6, 20, 21].

In this case, the trigger for EGID seems to have been dairy foods, as evidenced by his improvement with an elimination diet that specifically involved dairy avoidance. Of note, the patient most likely had remote toxocara exposure in the years prior to the development of primary EGID based on the finding of detectable toxocara antibody. Given that the toxocara antibody titer was low, and there was no detectable parasite on stool studies or histopathology, we felt he did not have evidence of active parasitic infection. In addition, while chronic pancreatitis may cause diarrheal symptoms, it would not explain the eosinophilic infiltrates in the GI tract, and he notably did not respond to pancreatic enzyme supplementation.

Our case raises the possibility of a link between prior parasite exposure and the future development of EGID, possibly via sensitization of adaptive immunity and subsequent cross-reactivity with specific food antigens (as evidenced by the effectiveness of the elimination diet) [22]. Parasite exposure as a risk factor for EGID should be investigated in larger cohorts. Additionally, this patient had been previously diagnosed with irritable bowel syndrome; this may have delayed the diagnosis of EGID as a result of misattribution of his symptoms during prior hospitalizations. Thus, this case also underscores the importance of exploring alternative diagnoses in patients with presumed irritable bowel syndrome who present with severe symptoms necessitating hospital admission.

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Compliance with ethical standards

Conflict of interest The authors declare no conflicts of interest or sources of funding associated with this article.

Human and animal rights No animals were utilized in the creation of this report. No humans were harmed in the creation of this report.

Informed patient Informed patient consent was obtained for publication of the case details.

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