A rare case of eosinophilic esophagitis and eosinophilic subserosal gastroenteritis with ascites

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Dear Editor,

Primary eosinophilic gastrointestinal disorders are defined as disorders that primarily affect the gastrointestinal tract, with eosinophils predominant inflammation in the absence of known causes for eosinophilia (e.g., drug reactions, parasitic infections, inflammatory bowel diseases, and malignancy). These disorders include eosinophilic esophagitis (EoE), eosinophilic gastroenteritis (EG), and eosinophilic colitis (1).

Here we present the case of a 30-year-old woman with a 2-month history of upper abdominal pain, nausea, vomiting, abdominal distension, and weight loss of 5 kg. Her laboratory test results on admission revealed peripheral eosinophilic leukocytosis with 67.7% eosinophils. Her serum IgE and tumor marker Ca-125 levels were elevated nearly twofold and tenfold, respectively. Serology for all types of viral hepatitis, Echinococcus granulosus, and trichinosis as well as stool examinations for parasites, ova, and other common pathogens were negative. Immunology tests, including screening for celiac disease, were negative. Diagnostic paracentesis showed a large number of eosinophilic granulocytes without malignant cells seen in punctate.

Abdominal computed tomography (CT) revealed increased thickening of the esophageal and small bowel wall with ascites (Figure 1). Upper endoscopy findings were normal except for marginally narrowed esophageal lumen, and at least four biopsy samples from each site were obtained from the upper and lower part of the esophagus, stomach, and duodenum. Histopathology showed esophageal mucosa pervaded by eosinophilic granulocytes (>15 in the high-power microscopic field (HPF) (Figure 2a). Histopathology of antral and duodenal biopsies was unremarkable.

The constellation of clinical presentation, histopathological and cytological findings, and CT findings were suggestive of EoE and subserosal EG. The patient was administered oral prednisone in a tapering manner. Subsequently, topical steroid fluticasone was introduced at a dose of 440 mcg twice daily and was tapered to 220 mcg twice daily after 8 weeks. Additionally, the patient was prescribed the six-food elimination diet, during which she excluded the six-most probable food allergens: milk and

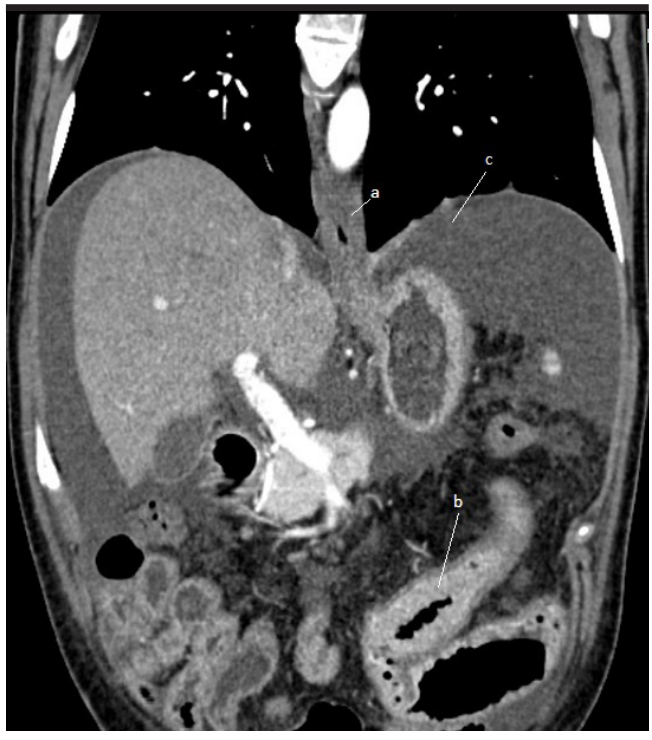

Figure 1. Abdominal CT with intravenous contrast demonstrated: a) circular thickening of the esophagus wall, b) thickening of the small bowel (jejunum), c) ascites in perisplenic, perihepatic, interintestinal area.

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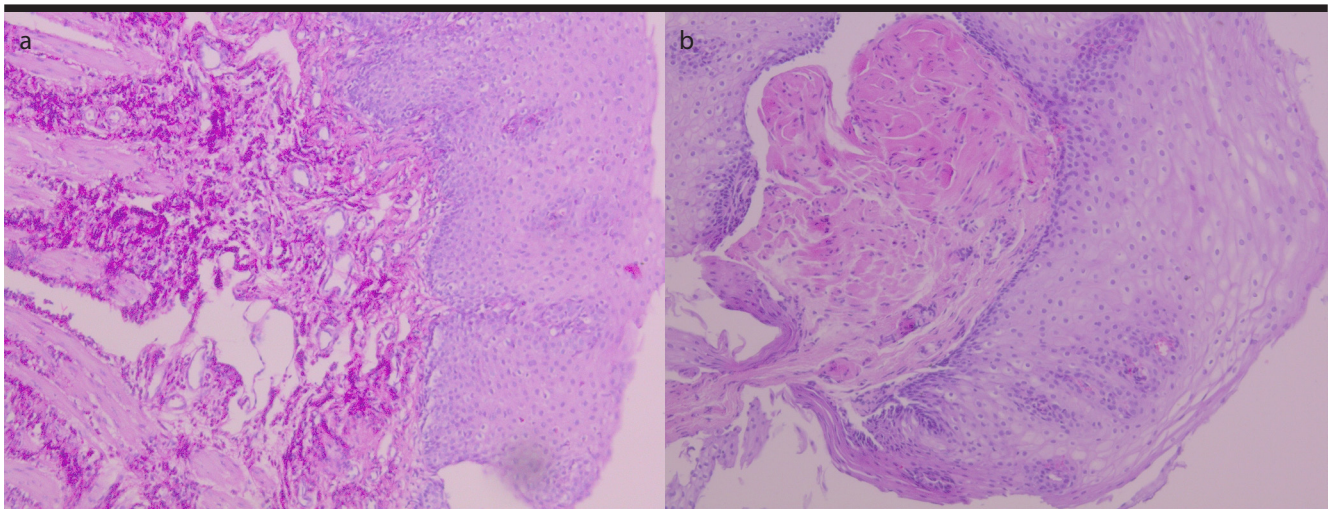

Figure 2. a, b. a) Eosinophilic esophagitis. On the surface regular squamous epithelium, and below mixed inflammatory infiltrate of lymphocytes and eosinophilic granulocytes. Hematoxylin and eosin magnification (HE, x100); b) Esophageal mucosa. On the surface regular sguamous epithelium, and below there is no inflammatory infiltrate of lymphocytes and eosinophilic granulocytes. Hematoxylin and eosin magnification (HE, x100).

dairy products, wheat, eggs, soy, tree nut/peanut, and sea food.

After 1 month, her abdominal pain and ascites completely disappeared and blood test results revealed a reduced absolute eosinophil count (25.1%) and normal serum IgE levels. However, on repeated esophageal biopsies 3 months later, esophageal histology still showed more than 15 EoE per HPF. This could be explained by the possibility of poor compliance or inadequate administration of topical steroids or by the possibility that the medicine dose was not high enough to induce histological remission. Recently published papers suggest that a high dose of fluticasone, 880 mcg twice daily, has a higher cure rate, both in adults and children (2). Thus, the patient was administered fluticasone 880 mcg twice daily, and after 2 months, control biopsies revealed a histologic response of less than 15 eosinophils per HPF in esophageal specimens (Figure 2b). At present, she is feeling well and we plan to gradually stop the medical treatment and diet, although there are no firm data regarding the duration of maintenance treatment (2).

According to the latest consensus, esophageal eosinophilia responsive to the proton pump inhibitor therapy is considered to be a part of EoE continuum rather than a separate entity. Histopathologically, EoE is defined as >15 eosinophils observed in the HPF in at least one biopsy sample (3).

Although there are no standards for the diagnosis of EG, the following criteria were suggested by Talley et al: presence of gastrointestinal symptoms, histological demonstration of eosinophilic infiltration in one or more areas of the gastrointestinal tract or presence of high eosinophil count in ascitic fluid, and no evidence of parasitic or extraintestinal disease (4). Three types of EG are defined according to the Klein classification: mucosal, muscular, and subserosal. Clinical findings vary depending on the layer involved. Patients with serosal type of EG frequently have ascites (5).

In conclusion, our patient had concomitant EoE and subserosal EG complicated by ascites, which is very rare according to available literature. Although the cause of disease remains unknown, the patient achieved clinical remission after she was started on steroid therapy, with rapid symptomatic improvement and disappearance of ascites. During the 6-month follow-up period, all findings were within the normal range, and the disease did not recur for the indicated period of time. Although this is a rare entity, it should be suspected in the presence of peripheral eosinophilia with occurrence of eosinophilic ascites, especially after excluding known causes that can lead to this condition.

Informed Consent: Informed consent was obtained from the patient who participated in this study.

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