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A Case of Postpartum Eosinophilic Gastroenteritis and Review of the Literature

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Key Words
Eosinophilic gastroenteritis · Pregnancy · Postpartum period · Atopy · Allergy

Abstract
Eosinophilic gastroenteritis (EG) is a rare disease of unknown etiology that can involve any area of the gastrointestinal (GI) tract. It can be classified into three major types: predominantly mucosal, muscularis, or subserosal form. Diagnosis of EG is confirmed after the exclusion of other disorders having similar features, such as parasitic infection, carcinoma, allergy, and autoimmune conditions such as Churg-Strauss disease. Correct diagnosis hinges on the presence of eosinophilic infiltration of one or more areas of the GI tract, without extraintestinal involvement. We present the case of a 30-year-old female with symptoms of EG 26 days after delivery. After corticosteroid and montelukast treatment for 2 weeks, all symptoms and objective clinical findings disappeared. Although numerous cases of this disorder have been described, to our knowledge this is the first case of postpartum EG. This case highlights the need to include this entity in the differential diagnosis of postpartum GI disorders.

Introduction

Eosinophilic gastroenteritis (EG) is an uncommon and benign condition characterized by eosinophilic tissue infiltration that may involve any part of the gastrointestinal (GI) tract. It was originally described by Kaijser in 1937 [1], and hundreds of cases have since been reported. EG can affect both children and adults, with a peak incidence during the third to fifth decades of life [2]. However, the real incidence is difficult to estimate because many cases remain undiagnosed or not reported, and an approximation is about 1 case per 100,000 patients [3]. The etiology of EG is still undetermined, but allergic mechanisms may have an important role in the pathogenesis, as a vast number of patients exhibit increased total IgE and food-specific IgE levels [4, 5]. In 40–50% of patients, it is associated with a history of asthma and allergies [6, 7].

EG belongs to a group of primary eosinophilic GI disorders (EGIDs), along with eosinophilic esophagitis, food protein-induced enterocolitis, and eosinophilic proctitis. EGIDs represent an eosinophil-rich inflammation of the GI tract in the absence of extraintestinal affection and known causes for eosinophilia [8]. According to the classification by Klein et al. [9], based on histopathological findings of eosinophilic infiltration, EG can be divided

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into three groups: mucosal, muscularis, and (sub)serosal form of disease, although more than one layer may be involved. Clinical manifestations vary greatly, depending on which layer and part of the GI tract are predominantly affected. The mucosal form mostly presents with abdominal pain, vomiting, diarrhea, anemia, and protein-losing enteropathy. Obstructive symptoms are typical symptoms of the muscularis form, while serosal forms may include ascites, bloating, possible peritonitis, and a high peripheral eosinophil count [3].

This paper reports a case of EG in the early postpartal period. This is a case not yet described in the literature, and the possible association and immunologic interactions between pregnancy and EG will be discussed.

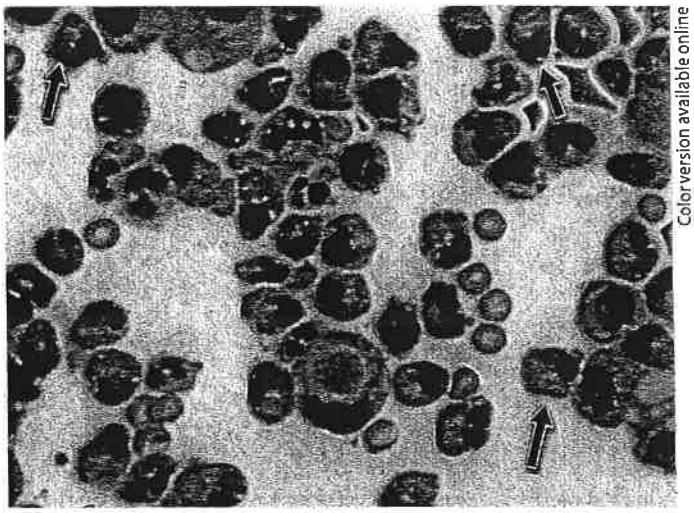

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Fig. 1. Cytological stains of ascitic fluid with arrows pointing at eosinophils.

Case Report

A 30-year-old female presented with a 14-day history of epigastric pain, vomiting, and diarrhea. Symptoms started 26 days after she gave birth to a healthy female child. She was regularly breastfeeding until the onset of symptoms, and there were no changes in clinical manifestations during the 14-day period. The patient had a history of atopy, including chronic rhinosinusitis and mild bronchial asthma, which began 3 years before. Her asthma was well controlled with use of low-dose inhaled glucocorticosteroids and rapid-acting β_2-agonists as needed. There were no documented food allergies. Physical examination showed abdominal distention and tenderness with clear signs of ascites. Possible gynecological causes were excluded by physical examination and transvaginal ultrasonography.

Laboratory evaluation revealed a slightly elevated white blood cell count of 11.2×10^9 (upper limit of normal 9.7×10^9) with 12% eosinophils (upper limit of normal 7%). The level of total IgE was elevated (129 kU/l; upper limit of normal 114 kU/l), with normal values of IgA (2.3 g/l; normal range 0.7–4.0 g/l). Hemoglobin, red blood cell count, electrolytes, creatinine, and liver function tests were within normal ranges. Stool analyses were negative for infective agents, as well as serological tests used to detect parasitic infestations as a possible cause of symptoms followed by eosinophilia. Anti-endomysial antibodies and tests for parasites were negative. We could not find any deterioration of C-reactive protein levels (1.2 mg/l; normal <5 mg/l), and rheumatoid factor, anti-endomysial, antinuclear, and perinuclear antineutrophil cytoplasmic antibodies were all negative. We performed paracentesis to obtain ascitic fluid. Biochemical and cytological analysis confirmed an exudative type of ascites with high protein amount (39 g/l), and a high cell count consisting predominantly of eosinophils (up to 40%) and phagocytes (fig. 1).

Because our patient suffered prolonged abdominal pain and vomiting, we performed upper GI endoscopy, revealing a normal esophageal, stomach, and duodenal mucosa. Lower endoscopy was the following procedure performed showing a macroscopically normal colonic mucosa. Biopsies from the esophagus, stomach, small intestine, and colon were taken, and histopathologic evaluation revealed a predominantly eosinophilic infiltration in all biopsy specimens involving the full thickness of GI mucosa through to the lamina propria (fig. 2). The density and depth of infiltration was similar in all the retrieved tissue. Stains for *Helicobacter pylori* were negative.

Based on our findings and taking into account the possible differential diagnosis, we concluded that it was a case of EG characterized by mucosal and serosal involvement. According to this diagnosis, we started treatment with 40 mg of prednisone and 10 mg montelukast daily. After 2 weeks of treatment, our patient was symptom free with no further relapses.

Discussion and Review of the Literature

EG is a rare disease presenting itself with nonspecific and various GI symptoms. Peripheral eosinophilia can be a distinguishable characteristic, and a history of allergies and atopy is another potential clue to the diagnosis; however, the eosinophil count is normal in about 25% of cases, with absence of allergic disorders in a patient's history in about 25–75% [3, 10]. The mucosal form of the disease is the most frequently diagnosed due to the possibility of endoscopic visualization of different mucosal changes and biopsy evaluation. Secondary causes of eosinophilic tissue infiltration, such as parasitic infestation, malignancy, allergic reactions, and autoimmune diseases (Churg-Strauss disease), as well as extraintestinal organ involvement should be excluded.

The etiology and pathogenesis of EG are still not well understood. The frequent concomitance of allergic disorders suggests that hypersensitivity reactions and eosino-

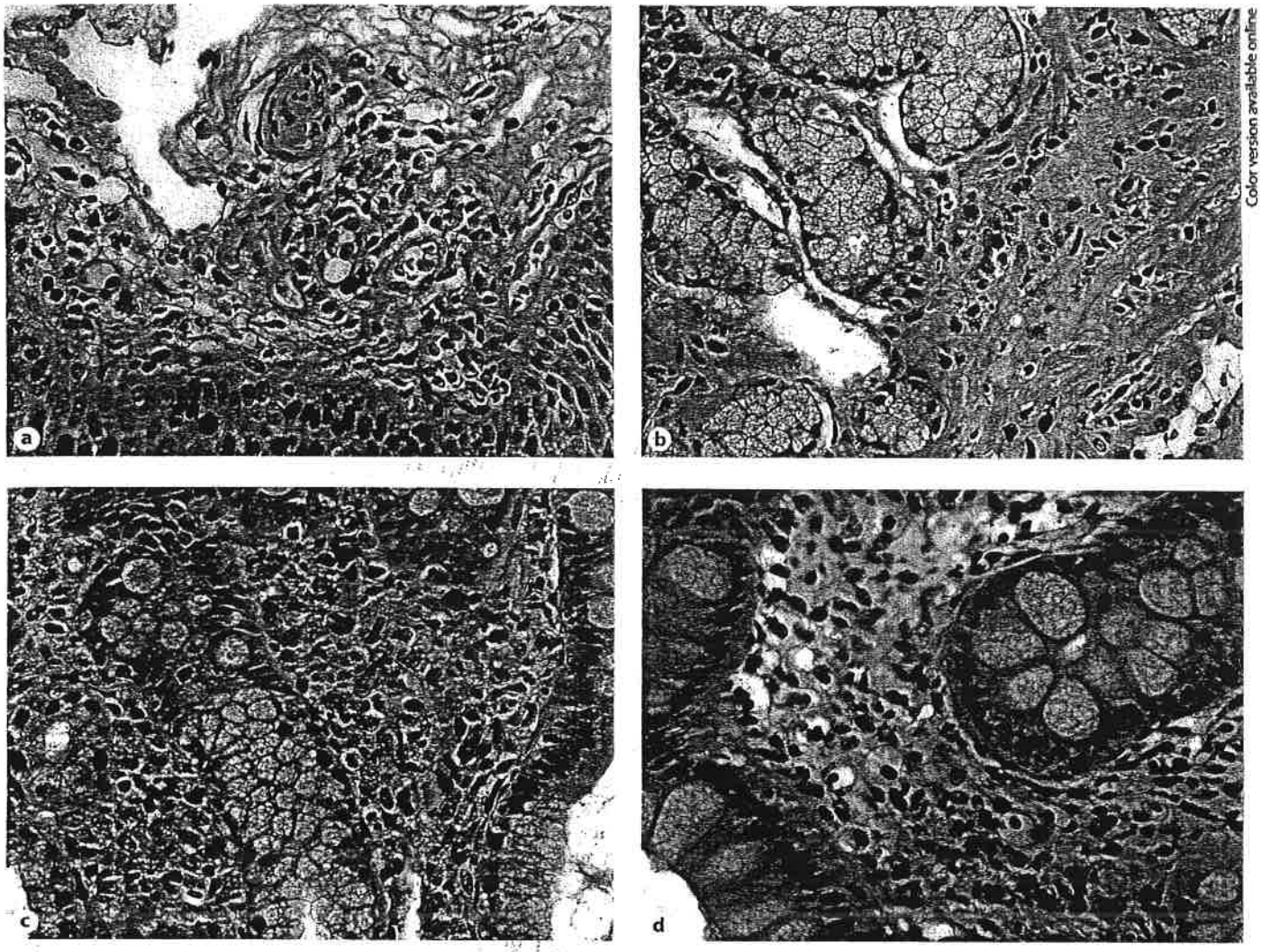

Fig. 2. Histopathology showing eosinophilic infiltration of mucosal layer through lamina propria. **a** Esophagus. **b** Stomach. **c** Duodenum. **d** Colon.

phils play a major role in the pathogenesis. Eosinophils are common residents in the lamina propria of the GI tract in the healthy state. The number of eosinophils that is regarded as pathological is still undetermined [11]. During eosinophilic inflammation of the GI tract, they function as antigen-presenting cells, and at the same time mediate cytotoxicity to the intestinal epithelium through various eosinophil-derived granule proteins [12, 13]. Apart from eosinophils, recent studies support the role of T helper 2 (Th2) allergic response and the production of cytokines (IL-3, IL-5, and IL-13), as well as chemokines such as eotaxin (eosinophil selective chemokine) [14–17]. The alteration of balance between Th1 and Th2 response caused by allergens in favor of Th2 hyperactivity and hyperproduction of Th2 cytokines is the most crucial factor in the pathogenesis of EG. Th2 cytokines in conjunction with eotaxin chemokines function as major inducers of tissue eosinophilia [6, 8]. Even though our patient had a history of allergic rhinosinusitis and asthma, it is more likely that in this case the immunological changes during pregnancy induced the above-mentioned mechanisms that resulted in disease manifestation. The predominant maternal immune response during pregnancy is humoral, which is why cell-mediated diseases, such as rheumatoid arthritis, are ameliorated during pregnancy, while other, such as systemic lupus erythematosus are aggravated. This is in accordance with a downregulated Th1-mediated immune response and an enhanced Th2-medi-

ated response [18, 19]. Therefore, it is possible that these changes during pregnancy have brought in our patient an outburst of EG in the early postpartum period, together with an existing atopy. Although more than 300 cases of EG have been described in the literature, our search found no results of EG during pregnancy or in the early postpartum period; neither has the connection between the two entities been discussed. This case highlights the need to include EG in the differential diagnoses of postpartum GI disorders.

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