

Eosinophilic gastroenteritis developed after remission of cow's milk allergy

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Key words eosinophilic gastroenteritis, food allergy, oral immunotherapy.

Eosinophilic gastrointestinal diseases (EGIDs) are characterized by abnormal eosinophilic infiltration of different segments of the gastrointestinal tract in the absence of identifiable secondary causes. EGIDs are often associated with other atopic conditions. Comorbid food allergies have been noted in only 3.4% of patients with eosinophilic gastroenteritis (EGE).¹ Although dietary treatments, including elimination diets and empirical food avoidance, are known treatment options for eosinophilic esophagitis, the effectiveness of dietary therapies for EGE remains to be clarified.² Since responses to dietary therapies do not always correlate with the specific food sensitization determined by skin test and/or serum antigen-specific IgE, it is difficult to identify a causative food for empirical avoidance. In this report we describe a case of EGE that developed after long-term sustained unresponsiveness of IgE-mediated cow's milk (CM) allergy that was effectively treated with the elimination of dairy products.

A female infant, exclusively breastfed from birth, experienced several episodes of urticaria within 1 h of ingesting CM and boiled egg during weaning. Because her serum-specific IgE (sIgE) (ImmunoCAP, Phadia, Uppsala, Sweden) to CM and egg white was 0.48 and 6.32 kUa/L, respectively, she was diagnosed as having IgE-mediated CM and egg allergies. Dairy and egg products were eliminated from her diet. At 2 years 10 months of age, a follow-up examination revealed decreases in sIgE. Dairy and egg products were reintroduced, and their intakes were gradually increased without elicitation of any symptoms. At 4 years 7 months of age, when viral enterocolitis was epidemic in the child's kindergarten, the child presented with vomiting and diarrhea. She was treated with *Bifidobacterium* probiotics. Four weeks later, she was referred to our hospital because of weight loss, persistent diarrhea, hematochezia, and a remarkable eosinophilia (WBC 24 700/ μ L, eosinophils 40%). On admission, serum sIgE showed; egg yolk 0.38 kUa/L, egg white 3.64 kUa/L, and CM 0.44 kUa/L. An allergen-specific lympho-proliferation index

against lactoferrin was positive, whereas those against κ -casein, and human α -lactalbumin were negative. Abdominal computed tomography revealed a thickened intestinal wall. Upper and lower endoscopic examinations showed no apparent mucosal lesions, but duodenum biopsy specimens obtained by endoscopy revealed infiltration of eosinophils (> 100 eosinophils/high-power field) in the mucosa (Fig 1). Eosinophil infiltration was not remarkable in gastric and ileal mucosa. Bone marrow examination showed no monoclonal cell proliferation, and *FIP1L1-PDGF α* and *JAK2* mutations were not detected. Parasite screening was negative. Chest computed tomography and echography did not show abnormal findings. The patient was diagnosed as having EGE. After 6 days of fasting and parenteral nutrition, her gastrointestinal symptoms disappeared and a diet excluding dairy and egg products was reinstated. Although egg products were resumed on the 18th day of hospitalization, absolute eosinophil counts continuously decreased until dairy products were reintroduced on the 25th hospital day. After commencement of dairy products, she complained of intermittent abdominal pains and presented with progressive diarrhea but no cutaneous or respiratory symptoms. Peripheral blood eosinophil counts increased over the following 5 days. These symptoms and eosinophilia disappeared after

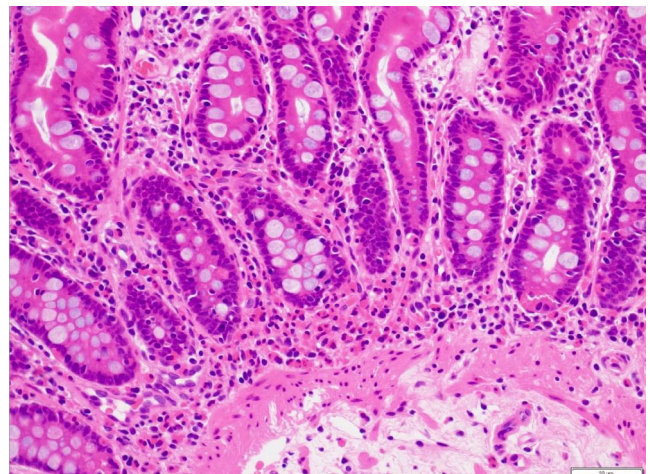


Fig. 1 Histologic findings in the duodenal mucosa. Numerous eosinophils (> 100/ high-power field) infiltrated the mucosa.

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Received 15 March 2019; revised 11 July 2019; accepted 13 September 2019.

doi: 10.1111/ped.14078

elimination of dairy products. Written informed consent to publish this report was obtained from the patient's parent.

This case indicates that CM antigens were involved in the pathogenesis of IgE-mediated food allergy and EGE in the same patient. Although her initial gastrointestinal symptoms could be due to eosinophilic gastroenteritis, the epidemic viral enterocolitis might have triggered the development of EGE after remission of IgE-mediated food allergy.

Eosinophilic gastroenteritis has been shown to occur in patients who underwent oral immunotherapy for IgE-mediated food allergy, and to remit following discontinuation of oral immunotherapy.³ Thus, increasing consumption of dairy products despite the presence of CM-sIgE might have induced EGE in the patient.

Inducible regulatory T cells (iTreg) have been demonstrated to play a critical role in sustained unresponsiveness and tolerance. iTregs lose their regulatory capabilities when present in inflammatory environments.⁴ Intestinal inflammation caused by viral infection could cause iTreg to lose their regulatory function or change their phenotype, resulting in a predominance of pathogenic Th2 cells.

In conclusion, pediatricians should be aware that the same food antigen can cause EGE after recovery from IgE-mediated food allergy. In cases with gastrointestinal symptoms associated with eosinophilia in children who recovered from IgE-mediated food allergy, food elimination and reintroduction of the previously sensitized food antigens should be performed in

order to confirm EGE and to identify the causative food for empirical avoidance as a dietary treatment.

Disclosure

The authors declare no conflicts of interest.

Author contributions

N.I. wrote the manuscript. Y.O. reviewed and supervised the manuscript. N.I., H.M., A.K., K.S. and Y.O. treated the patient. All authors read and approved the final manuscript.

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Indocyanine green lymphography for congenital lymphatic dysplasia with tuberous sclerosis complex: A case report

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Key words congenital lymphedema, extremely low birthweight infant, indocyanine green lymphography, lymphatic dysplasia, tuberous sclerosis complex.

Tuberous sclerosis complex (TSC) is an autosomal-dominant, multisystem disorder characterized by benign hamartomas in multiple organ systems. Congenital lymphedema has been described as a possible rare association of TSC.¹ However, there have been very limited reports of imaging evaluation of this clinical condition,¹ and no report of imaging study

regarding lymphedema in infants with TSC. We evaluated lymphatic function in an extremely low birthweight infant with TSC, using indocyanine green (ICG) lymphography to diagnose and assess the severity of lymphatic dysplasia.

A male neonate with a birthweight of 854 g, born by cesarean section after 25 weeks of pregnancy, was admitted immediately after birth to the neonatal intensive care unit. Intestinal perforation occurred on day 6, and he was transferred to our hospital for surgery. Cardiac ultrasonic examination before surgery revealed multiple cardiac intraventricular masses (Fig. 1a), and TSC was suspected. His diagnosis of TSC was confirmed clinically based on the findings of brain magnetic

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Received 17 November 2018; revised 22 August 2019; accepted 14 November 2019.

doi: 10.1111/ped.14071