

# A Case of Eosinophilic Esophagitis With Food Hypersensitivity

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Eosinophilic esophagitis (EoE) is a disorder characterized by isolated eosinophilic infiltration of the esophagus with esophageal symptoms. Although some patients with EoE are related to food hypersensitivity, it is hard to identify causative foods. This report describes a case of EoE with dysphagia. A 28-year-old man presented with dysphagia and substernal discomfort for 15 days. He had taken a protein complex for 2 months. Endoscopy showed several linear furrows and multiple mucosal nodularities on the lower and mid esophagus, and the biopsies of esophagus revealed marked eosinophil infiltration in the mucosa. The skin testing for the protein complex was positive. The patient was successfully treated with withholding treatment.

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## Key Words

Eosinophils, Esophagitis, Food hypersensitivity, Withholding treatment

## Introduction

Eosinophilic esophagitis (EoE) is a disorder characterized by isolated eosinophilic infiltration in esophageal mucosa. It was generally diagnosed to children, rarely occurred to adults. However, recently EoE is rapidly increasing in adults.<sup>1</sup> Since the first case was reported in 2002,<sup>2</sup> several cases have reported after 2008 in Korea.<sup>3,4</sup> In a single center study of 108 Korean patients who had dysphagia and refluxed symptoms, 1.8% of those patients were afflicted with EoE.<sup>5</sup> Another study informed that 0.4% of patients were diagnosed as EoE among 1,609 people

who underwent biopsies of esophageal mucosa.<sup>6</sup> Both reports shows that the EoE is a disease not just for western population but also for Asian including Korean.<sup>6</sup>

The cause of EoE is not well understood. However, most patients with EoE have allergic disorders such as food allergy, atopic dermatitis, asthma or allergic rhinitis,<sup>7</sup> and improve by corticosteroid treatment.<sup>8</sup> So it is predicted that the EoE is highly related to allergy. Through the withholding treatments based on the amino acid based formula and 6 most common allergen elimination diet after allergic evaluation, clinical and histological improvement was made in 50% to 90% of patients, which resulting food allergens are primary causes of EoE.<sup>9-11</sup> However, the causative

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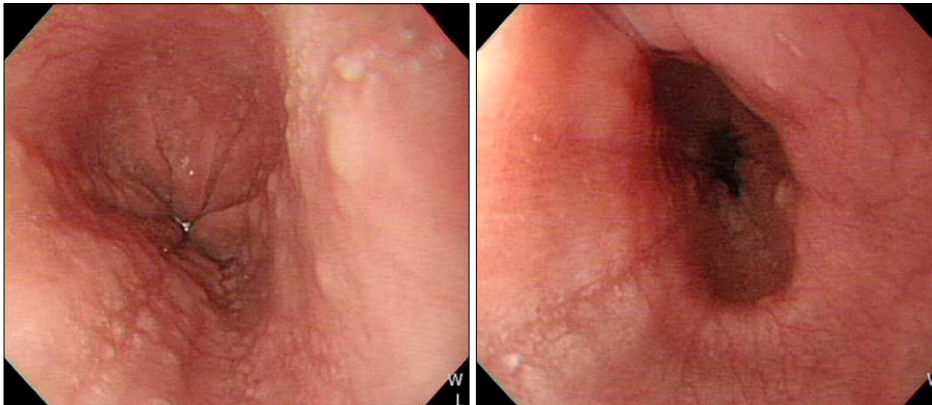
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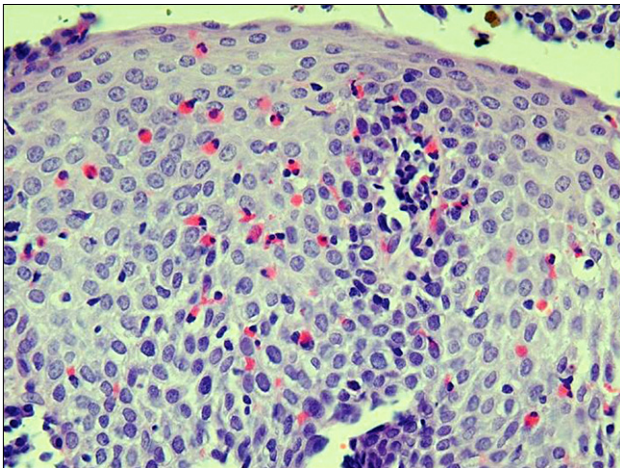
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**Figure 1.** Endoscopy shows some linear furrows and multiple mucosal nodularities on the lower and mid esophagus.



**Figure 2.** Microscopic finding of the esophagus shows a heavy eosinophilic infiltration, > 20 eosinophils per high power field (H&E,  $\times 400$ ).

foods were not identified or hard to find.

We experienced a case of EoE with hypersensitivity which was induced by specific food identified through the skin testing, and improved by withholding target food.

## Case Report

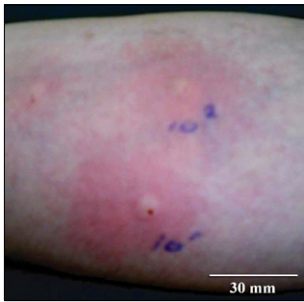
A 28-year-old man visited gastrointestinal clinic because of dysphagia, nausea, and substernal discomfort for 15 days. He did not have any past allergic histories and family histories of an atopic disease. Physical examination was unremarkable, and any skin lesions were not observed. The laboratory testing showed a white blood cell count of  $6,470/\text{mm}^3$  (eosinophils 4.3%) and a total IgE level of 147 IU/mL (Class 3). It is significant as allergen when

the class values of total IgE are 2 or above.

Esophagogastroduodenoscopy showed some linear furrows and multiple mucosal nodularities on the lower and mid esophagus without reflux esophagitis (Fig. 1), and we took 3 specimens at the lower and mid esophagus, respectively, under suspecting EoE. We also performed multiple biopsies at the stomach and bulb of the duodenum. Whereas multiple mucosal biopsies on the stomach and duodenum were normal, heavy eosinophilic infiltration was observed on the esophageal mucosa (Fig. 2). Based on the clinical, endoscopic and histological findings, the patient was diagnosed as EoE. We found out he had eaten the food (protein complex; Myoplex<sup>®</sup>, Abbott Laboratories, Abbott Park, Illinois, USA) for 2 months while he built his body. Allergic evaluations for identifying causative allergens were performed by consulting with the specialist of the allergic department. Multiple allergen simultaneous tests showed a hypersensitivity to peanut. Also, we confirmed a hypersensitivity to the protein complex (Myoplex<sup>®</sup>) by intradermal injection test (Fig. 3). We first started treatment with proton pump inhibitors after endoscopic examination, but the symptoms were improved partially after 2 weeks. After we confirmed the result of biopsies, withholding treatment was added. The patient's symptoms were improved gradually since the withholding treatment. Two months later, follow-up endoscopy revealed a disappearance of linear furrows and mucosal irregularities, and mucosal biopsies showed marked decrease in eosinophil counts less than 5 eosinophils per high power field. The patient was stable without recurrence at the 1 year follow-up.

## Discussion

EoE is characterized by (1) esophageal symptoms such as dysphagia, food impaction, or regurgitation; (2) eosinophilic in-



Myoplex <sup>®</sup>	Wheal size (mm)	Erythema size (mm)
1:100	7 x 5	30 x 30
1:10	7 x 5	35 x 40
Histamine (control)	4 x 4	20 x 15

**Figure 3.** Intradermal skin tests with protein complex (Myoplex<sup>®</sup>) represent hypersensitivity to the dilution of protein complex compared to histamine. The size (mm) of wheal and erythema by Myoplex<sup>®</sup> are bigger than those by histamine.

filtration at esophageal mucosa ( $\geq 15$  eosinophils per high power field) and (3) unresponsiveness to high-dose proton pump inhibitors.<sup>12</sup> EoE was thought to be a common disorder in children, but recently it also has become more prevalent in adults. Although the pathogenesis of EoE is still not completely understood, it has been thought to relate to allergic disorders, especially food hypersensitivities.<sup>7</sup>

Food hypersensitivity reactions are divided into IgE-mediated, non-IgE-mediated and mixed ones. Whereas IgE-mediated reactions are usually immediate and mainly involve the skin, non-IgE-mediated reactions are delayed or chronic and predominantly manifest in the gastrointestinal tract and skin. IgE-mediated reactions can be confirmed by skin prick test and food specific IgE levels in the serum. Also, atopy patch tests are being used to detect delayed, T-cell-mediated hypersensitivity. EoE is thought to be a mixed-reaction disorder, and the combination of skin prick test and atopy patch test may be more effective to identify the causative foods.<sup>13</sup>

Therapies of EoE include drug (eg, corticosteroid) and diet treatment.<sup>12</sup> Although the use of systemic or topical steroids improves clinical symptoms effectively, EoE recurs in more than 90% of patients after treatment is completed.<sup>14</sup> Diet treatment can be divided into 2 categories: amino acid-based formula and elimination diet. Kelly et al<sup>9</sup> reported the effectiveness of an amino acid-based formula in 10 pediatric EoE patients. Spergel et al<sup>13</sup> demonstrated that 26 children with EoE improved in both symptoms and esophageal inflammation by dietary elimination of foods. Also, they recommended skin prick and atopy patch tests to identify causative foods.<sup>11</sup> Kagalwalla et al<sup>10</sup> also examined 60 children with EoE by a 6-food elimination diet (cow's milk, soy, wheat, egg, peanut and seafood) or an amino acid-based formula. Clinical and histological improvements were observed in 74% and 88%, respectively. In adults, however, the evidence of link between food hypersensitivity and EoE was limited. In a recent study, 81% of adults with EoE had  $\geq 1$  allergens identified by

skin prick testing and 50% had  $\geq 1$  skin tests positive to food allergens.<sup>15</sup> The most common food allergens consisted of peanut, egg white, soybean, milk and tree nuts in their study. In our case, we suspected causative food (consist of cow's milk, egg and wheat) by history taking, confirmed it by skin testing, and achieved clinical, endoscopic and histological improvements after avoiding the food. Therefore, allergic evaluations for identifying food triggers should be recommended to all adults with EoE.

In conclusion, the link between food hypersensitivity and EoE has not been well established and there are controversies about the effectiveness of diet treatment in adult patients with EoE. Herein, we experienced a case of EoE which was induced by specific food identified through the skin testing, and improved by withholding target food. More studies are needed to demonstrate the effectiveness of diet therapy in adult patients with EoE.

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