



Letter

Eosinophilic esophagitis to unsuspected rare food allergen

Introduction

According to the International Gastrointestinal Eosinophilic Researchers Consensus, “Eosinophilic esophagitis (EoE) represents a chronic, immune/antigen mediated, esophageal disease characterized clinically by symptoms related to esophageal dysfunction and histologically by eosinophil-predominant inflammation.”¹ The onset is usually in children and young adults. The presenting symptoms depend on the patient's age and may include feeding difficulty, vomiting, abdominal pain, dysphagia, and in severe cases, food impaction.¹ Adult patients also may complain of heartburn, cough, or choking. The symptoms can be vague and misdiagnosed, often as gastroesophageal reflux disease, with a prolonged course and complications. The correct diagnosis may not be reached until esophageal stricture develops. The diagnosis is based on esophageal endoscopy and biopsy examination revealing at least 15 eosinophils per high-power field.¹

In some patients, food hypersensitivity is involved and can be suspected by the medical history or by skin testing (prick or patch) or serum specific immunoglobulin E (IgE) testing. A trial of elimination of the most common causative foods (milk, egg, wheat, soy, peanut/tree nuts, and fish/seafood) without testing has been recently advocated, to be followed by re-introducing those foods one by one to identify the causative food.^{2–4}

We present a case of an adult in whom the causative food would not have been identified by a trial of an empiric elimination diet or by testing for common foods only.

Case Report

A 58-year-old white man, a university professor, had a history of upper gastrointestinal symptoms for more than 15 years, which had been treated as gastroesophageal reflux disease. He also had allergic rhinitis (treated with antihistamines when needed) and asthma (controlled with inhaled corticosteroids), autoimmune alopecia, and splenectomy for autoimmune thrombocytopenia at 22 years of age.

He was initially treated with H₂ receptor blockers and then proton-pump inhibitors, with only partial improvement. During the subsequent 6 years, symptoms worsened and reached significant dysphagia. Esophagogastroduodenoscopy showed mild V-line longitudinal furrow in the distal esophagus and reflux esophagitis at the gastroesophageal junction. Results of esophageal biopsies revealed marked esophagitis with increased eosinophils; on average greater than 60/high-power field (hpf; Fig 1A), confirming the diagnosis of EoE. Duodenal and stomach biopsy

specimens showed mild to moderate chronic gastritis, but no evidence of celiac sprue or *Helicobacter pylori*. Swallowed viscous budesonide 0.5 mg daily was prescribed by the gastroenterologist, but it was taken irregularly. Over the next 2 years, he had increased nausea, dyspepsia, and dysphagia. A second esophagogastroduodenoscopy revealed normal appearance of the esophageal, gastric, and duodenal mucosa, but the esophageal mucosal biopsy showed 0 to 17 eosinophils/hpf (Fig 1B). The patient was then referred to our allergy/immunology clinic for food hypersensitivity evaluation.

The medical history did not reveal any suspected foods. Total serum IgE was elevated (499 KU/L), and specific IgE was only slightly positive to wheat (0.56 KU/L) and negative to milk and egg. His white blood count was normal with moderate eosinophilia (7.6%; 650/ μ L). Skin prick testing with 44 food extracts (ALK-Abello, Round Rock, TX) was positive (>3-mm wheal greater than the negative control) only to garlic and cottonseed extracts, which were not close to each other or to the histamine control. On revealing the results to the patient, he reported frequent eating of garlic-containing foods but not of cottonseed-containing foods (donuts, chewy candy, or pastries). We recommended a trial of strict avoidance of foods containing garlic and cottonseed and regular intake of budesonide 0.5 mg twice daily as viscous in sucralose powder (Splenda) between meals. Within a few weeks, he reported marked improvement, and after 3 months, budesonide was reduced to once daily, without symptoms. He has been asymptomatic for more than 2 years so far, except on a couple of occasions when he consumed a garlic-containing food. Recently, while on swallowed budesonide and inhaled corticosteroids for asthma, he developed oral candidiasis and was treated with voriconazole after no response to fluconazole. Interestingly, he had only minimal symptoms for 1 day during a 10-day period of discontinuation of budesonide. He felt better by taking 1 daily maintenance dose of budesonide, particularly with the possibility of accidental exposure to garlic. Oropharyngeal rinsing after swallowed budesonide or inhaled steroid was emphasized.

Discussion

Foods commonly reported to be implicated in EoE in children include milk, egg, and wheat, followed by beef, soy, and chicken.⁵ In adults, the most common foods are legumes, nuts, fruits, wheat, and milk, followed by soy and egg.^{6,7} Because testing for the causative food is frequently unrevealing, a trial of an empiric “6-food elimination diet” has been recently advocated^{2,6,7} to be followed by reintroduction of the foods individually. This prolonged approach not only deprives patients from numerous common foods, but can also miss other food allergens. EoE patients can be sensitive to rare foods, such as pea, mustard,

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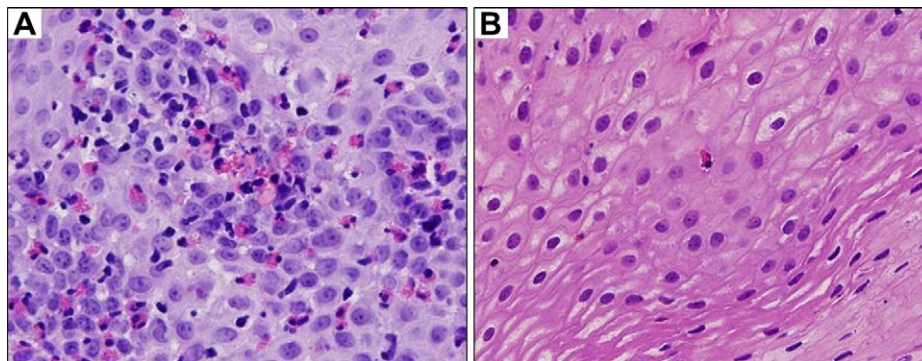


Figure 1. Esophageal mucosal biopsy showing remarkable eosinophilic infiltration before swallowed budesonide treatment (A) and markedly decreased eosinophils 2 years after treatment with swallowed budesonide (B).

carrot, almonds, string beans, pork, broccoli, rye, and corn.⁸ In a series of 26 subjects, only 1 patient had positive skin prick test to garlic among many other foods, but the clinical relevance to EoE was not confirmed.⁹

Our case exemplifies multiple interesting issues. First, the patient's initial symptoms were diagnosed as gastroesophageal reflux disease, but he had only partial improvement on proton-pump inhibitors therapy. Yet, evaluation for EoE was delayed for many years. Second, a thorough medical history did not reveal any suspected food; yet, skin prick testing with 44 food extracts was positive to garlic and cottonseed, which we initially did not think clinically relevant until the patient admitted his frequent consumption of garlic-containing foods. This demonstrates the crucial role of performing skin testing in this case. Third, his esophageal mucosa appeared healthy on endoscopy; yet, a biopsy revealed eosinophils. Hence, a normal appearance of the mucosa should not deter from a biopsy examination.¹ Fourth, garlic as a spice would not have been identified if the evaluation was by the "six-food elimination diet."^{2–4} Fifth, the common practice of testing (skin prick testing, patch, or specific IgE) to a limited number of foods would not have identified the culprit, particularly because no food was suspected by the history. Our patient may be the first documented case of garlic-related EoE. With the increasing use of various spices, attention should be given to such ingredients that are often hidden or unsuspected by patients.¹⁰

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