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Caustic Ingestion: A Possible Cause of Eosinophilic Esophagitis?

abstract

Eosinophilic esophagitis (EoE) is an emerging disease in both pediatric and adult patients. It is a chronic disease of the esophagus and refers to intense eosinophilic infiltration limited to the esophageal epithelium in the absence of gastroesophageal reflux disease. In most patients, EoE is thought to be part of an allergic response to food antigens or aeroallergens. One such trigger could be caustic damage of the mucosa. To the best of our knowledge, the following case report describes for the first time the possible association between caustic injury of the esophagus and EoE. *Pediatrics* 2013;131:e1284–e1287

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KEY WORDS

eosinophilic esophagitis, eosinophil granulocyte, children

ABBREVIATIONS

EoE—eosinophilic esophagitis

GERD—gastroesophageal reflux disease

HPF—high-power field

PPI—proton pump inhibitor

Dr Homan conceptualized and designed the case report, and drafted the initial manuscript; Dr Orel reviewed and revised the manuscript; Dr Liacouras critically reviewed and revised the manuscript; and all authors approved the final manuscript as submitted.

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Eosinophilic esophagitis (EoE) is an increasingly diagnosed disease especially in the developed world.¹ It represents a chronic, immune/antigen-mediated esophageal disease characterized clinically by symptoms related to esophageal dysfunction and histologically by eosinophil-predominant inflammation.² The mechanisms underlying recruitment of eosinophils to the esophageal mucosa are not completely understood. The eosinophilic inflammation in patients with EoE is almost always triggered by dietary proteins, less likely aeroallergens.³ In a minority of cases, other triggering factors can be involved, such as viruses, helminthes, and tissue injury.⁴ The following case report describes a patient who developed EoE after a caustic ingestion. To the best of our knowledge, this is the first report of possible association between caustic injury of the esophagus and EoE.

CASE REPORT

An 8-year-old boy presented to our unit after accidental ingestion of strong acid

in a liquid form. His family history was strongly positive for allergic diseases and negative for EoE or symptoms typical for EoE. His mother had asthma, his father and sister had atopic dermatitis. His previous history was not contributory. He had no history of any kind of allergic diseases. In addition, he did not describe any gastrointestinal problems. His mother worked in a cleaning service and brought home lime scale remover (pH 1). She stored it in a bottle normally used for juice. The patient mistakenly ingested a few swallows and stopped because the liquid had a bad taste. Soon afterward, he began drooling and developed retrosternal pain after drinking water and was taken to the emergency care unit. On an initial clinical examination, ulcers on the base of tongue were detected and he was admitted to the hospital.

Endoscopy was performed a few hours later and revealed noncircumferential fibrin exudates and erythema in the upper half of the esophagus (Fig 1). The rest of the esophageal, stomach, and

duodenal mucosa appeared normal. The biopsies on initial endoscopy were not taken because of concern of possible perforation. He was treated with a proton pump inhibitor (PPI) in a dose of 1 mg/kg, antibiotics, and liquid diet, and was discharged 7 days later in good condition. We prolonged the PPI therapy for 3 months.

He returned to the outpatient clinic 1.5 years later because of swallowing problems that started 6 months before his visit. He also reported retrosternal pain after swallowing solid food. A repeat endoscopy demonstrated multiple white plaques throughout the esophagus. *Candida esophagitis* was suspected and he was treated with an antifungal medication. Eosinophils were prominent in the esophageal biopsy specimens (mean eosinophil count of 37.5 eosinophils per high-power field [HPF] and peak eosinophil count of 50 eosinophils/HPF). Eosinophilic microabscesses were also described. *Candida albicans* was not found. Antifungal treatment was discontinued and PPIs in a dose of 2 mg/kg were begun.

A third endoscopy was performed 2 months later (Fig 2). It continued to reveal full-length white speckles along the esophagus. Despite treatment with PPIs, the histology of the esophagus showed moderate inflammation with prominent esophageal eosinophilic infiltrate (mean eosinophil count of 22.7 eosinophils/HPF and peak eosinophil count of 30 eosinophils/HPF). Laboratory testing revealed an elevated serum immunoglobulin E level, positive skin-prick tests for soya and nuts, and positive patch test for soya. We introduced the specific hypoallergenic diet and performed another endoscopy 2 months later. The result was the same as with the previous endoscopy: white speckles along the whole esophagus and intense eosinophilic infiltration (mean eosinophil count of 34.8 eosinophils/HPF and peak eosinophil count of 50 eosinophils/HPF),

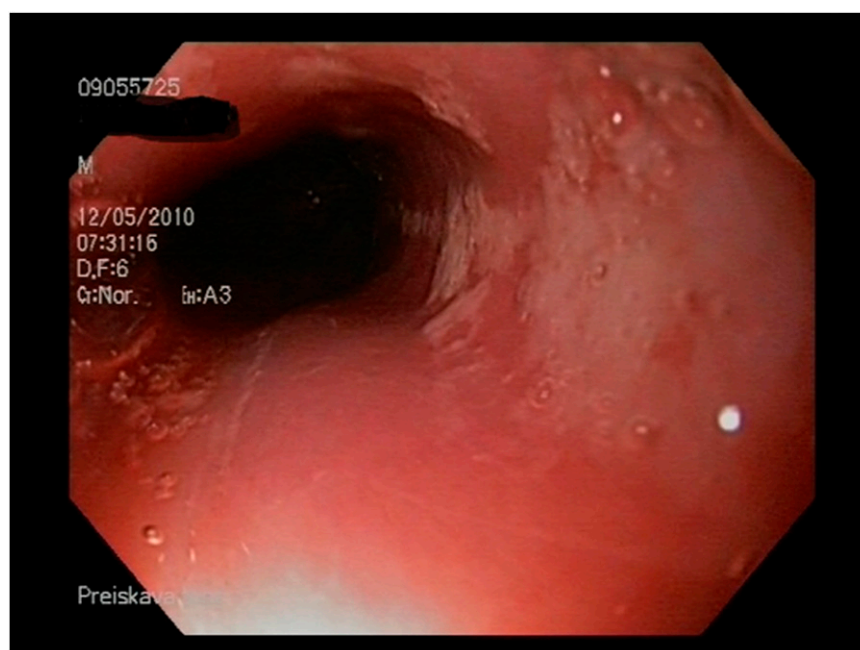


FIGURE 1
Noncircumferential fibrin exudates in the upper half of the esophagus.



FIGURE 2
White speckles along the esophagus.

so we introduced treatment with oral viscous budesonide.

DISCUSSION

Our report identifies a boy with predisposing factors for EoE and with no esophageal or gastrointestinal symptoms until he had a caustic acid ingestion. Immediately after the ingestion, typical changes for caustic injury of oral and esophageal mucosa were appreciated. He was treated for a caustic ingestion and his symptoms resolved. He remained asymptomatic for more than 1 year and then developed dysphagia and symptoms of gastroesophageal reflux. Subsequently, repeat endoscopies did not reveal an esophageal stricture, typically seen in patients with caustic ingestions, but instead white plaques and eosinophilic inflammation. Typically, the esophageal complications of a caustic ingestion occur within days or weeks of the ingestion. Delayed complications almost always consist of esophageal strictures. When he developed additional symptoms, these symptoms did not occur until more than

1 year after initial presentation and these later changes were diffuse. In addition, a repeat histology evaluation of esophageal mucosa after PPI treatment confirmed important eosinophilic infiltration unresponsive to acid suppression and suggested that the caustic ingestion may have been a precursor for the development of EoE in our patient.

Caustic injuries of the digestive tract are quite uncommon and the symptoms and complications are well known. EoE is becoming an increasingly recognized disease, with an increase in incidence and prevalence.⁵ Caustic ingestion injuries typically cause immediate neutrophilic esophageal inflammation. Acid ingestion also typically causes a gastric outlet obstruction, which did not occur in our patient. In contrast, EoE causes an isolated esophageal eosinophilia. Although it is possible that our patient had EoE before the caustic ingestion, we do not believe that this was the case, as he had no previous history of dysphagia, heartburn, re-

gurgitation vomiting, or retrosternal pain.

We propose 2 possible mechanisms for the development of EoE in our patient. First, it is possible that the caustic ingestion primarily damaged the esophageal mucosa and triggered pleiotropic eosinophils to migrate to esophageal mucosa and cause inflammation resulting in EoE. However, for this to have occurred, we would have expected that it would not have taken more than a year for the patient to have a recurrence of symptoms. Second, it is possible that breakdown of the esophageal mucosa barrier by the caustic material allowed EoE to develop by enabling food antigens to trigger an antigen/immune response that led to EoE. This finding is important, as the mechanism behind a caustic ingestion triggering EoE could be applied to individuals with severe grades of gastroesophageal reflux disease (GERD). Although previous reports have demonstrated that EoE can occur without the presence of GERD, our case may suggest that patients with severe grades of GERD, use of PPIs, or caustic damage may be more susceptible to developing EoE.⁶

Other possible associations and triggers have been reported, including diseases such as celiac disease,⁷ Rubinstein-Taybi syndrome,⁸ and autism spectrum disorder.⁹ Squires et al¹⁰ reported on 3 patients with a possible role of herpes simplex virus infection in developing EoE. The second case of the series was a 16-year-old boy with previous caustic damage. The description of this patient was vague and it was difficult to determine if the development of EoE in this patient was associated with a herpes infection or a past caustic ingestion. The sequence of events in our patient strongly suggests that the caustic ingestion was a precursor for the development of EoE. We conclude that EoE may be more likely to occur in patients with previous caustic damage.

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