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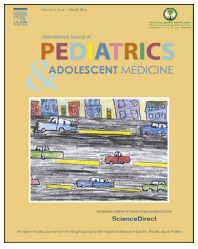

CASE REPORT

Esophageal perforation: An uncommon initial manifestation of eosinophilic esophagitis

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Abstract EoE-Perforation: Eosinophilic esophagitis (EoE) is commonly observed in children and young adults. Common manifestations of EoE include dysphagia and food impaction in adolescents and adults, whereas children present with failure to thrive, regurgitation or heartburn and abdominal pain. We describe two patients presenting with esophageal perforation and EoE. Diagnosing perforation promptly is critical to minimize and/or to avoid the multitude of complications resulting from esophageal perforation and to treat EoE because if left untreated, this condition may result in the recurrence of perforation or major morbidity and, rarely, death.

1. Introduction

Eosinophilic esophagitis (EoE), an immune-mediated disorder, is diagnosed by the combination of eosinophilic infiltration of the esophagus and esophageal dysfunction [1,2]. EoE is an increasingly recognized disease with a prevalence of 1–5 per 100,000 persons in the USA and Europe [2]. As a 'newer' disease, its natural history remains unclear; however, evidence has shown that EoE is not a premalignant condition and that the disease course

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varies from a prolonged spontaneous remission with or without treatment to a waxing and waning course; it can also be progressive, leading to strictures [2]. EoE usually presents insidiously with feeding difficulty, abdominal pain, gastroesophageal reflux-like symptoms, and dysphagia or acutely with food impaction. A rare and dramatic initial manifestation of EoE is esophageal perforation. We describe two adolescent males who presented with esophageal perforation associated with eosinophilic esophagitis.

2. Patient #1

L.C. was a 10-year-old boy who presented in December 2010 with chest pain and a fever of 103 °F for the duration of two weeks. He had no difficulty breathing, pain on inspiration, breathlessness, trauma or history of illicit drug or alcohol use. Past medical history included mild intermittent asthma and intermittent dysphagia for solid food for approximately one year. He had no prior esophageal or gastric surgery. On physical examination, his weight, height and vital signs were normal. His lungs were clear to auscultation bilaterally, and he had no abdominal tenderness or masses. No crepitations were found in the neck or chest. His WBC count was 11,800/ml, his hemoglobin level was 12.8 gm/dl and his platelets were 392,000/ml. The absolute eosinophil count was 440/ml, and the chest X-ray was normal. A computed tomography (CT) of the chest showed esophageal perforation with extra fluid and gas in the mediastinum adjacent to the esophagus (Fig. 1). The thoracic portion of the esophageal wall was thickened. An esophagram confirmed a contained esophageal perforation (Fig. 2). He was initially treated with intravenous antibiotics, and no drugs were administered orally. After one week, his symptoms improved, and he was started on omeprazole at 40 mg/day. Two weeks later, the follow-up CT scan was normal. At two months later, upper gastrointestinal endoscopy showed linear furrows and multiple white exudates throughout the esophagus and a small diverticulum in the midesophagus (site of perforation). Biopsies noted 23 eosinophils/HPF in the distal esophagus and 26/HPF (normal-usually none) in the proximal esophagus, along with marked basal cell hyperplasia and eosinophilic microabscesses. Gastric antral and duodenal biopsies were normal. He was started on topical fluticasone 440 mcg, four times a day, for 8 weeks. He was non-adherent to the treatment regimen and was lost for follow up. He presented again two years later with chest pain and fever as before and had similar findings on examination. An esophagram showed a contained perforation at the same site where an outpouching was previously observed. He had medical management as before, and his symptoms, including fever, improved within a week. His follow up esophagram was normal. Additionally, he had two follow up endoscopies, and the last one, three years after the initial presentation, showed mild furrows and a few white spots, with esophageal eosinophil count as 50—75/HPF in the distal esophagus and 25—45/HPF in the midesophagus. Allergy evaluation was recommended but was not followed through by the patient. Despite counseling, the patient continued to be non-adherent and had waxing and waning dysphagia but no food impaction, perforation, heartburn, regurgitation or chest pain. Growth continued normally.

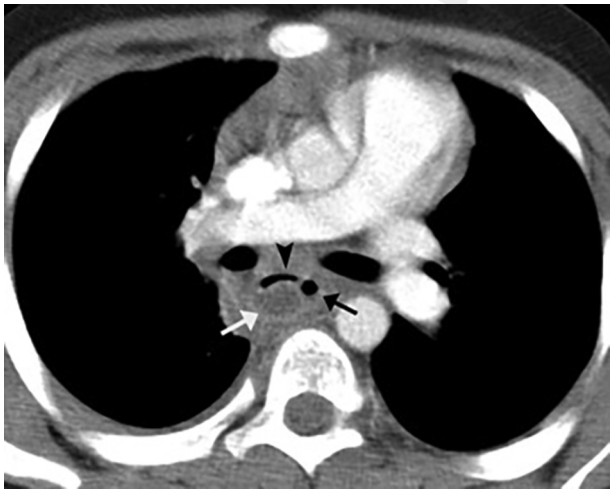

Figure 1 CT scan of the chest shows evidence of esophageal perforation, including a mediastinal fluid collection (white arrow) and extraluminal gas (black arrow). The fluid collection causes anterior displacement and narrowing of the esophagus (black arrowhead).

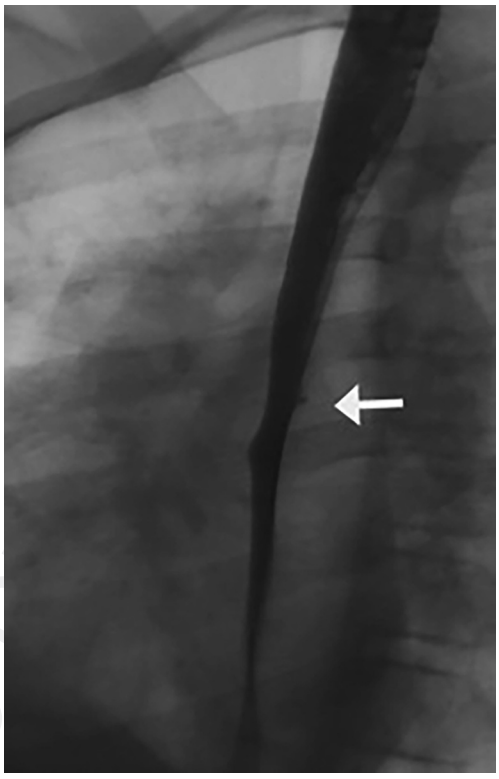

Figure 2 Esophagram performed with water soluble contrast material shows a small outpouching from posterior wall of esophagus indicating a contained esophageal perforation. Diffuse narrowing of a long segment of the mid esophagus is consistent with underlying eosinophilic esophagitis.

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