Transmural Eosinophilic Infiltration and Fibrosis in a Patient With Non-Traumatic Boerhaave's Syndrome Due to Eosinophilic Esophagitis

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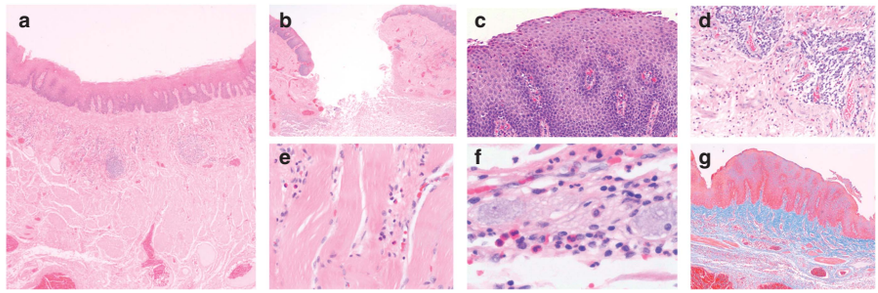

Figure 1. Histopathological evaluation of the entire esophageal wall in a patient with eosinophilic esophagitis presenting by Boerhaave's syndrome. Eosinophils permeate the submucosal and muscle layers and the neuronal plexus, promoting fibrous remodeling, intense collagen deposition, and smooth muscle hypertrophy (see detailed explanation in the text).

To the Editor: Eosinophilic esophagitis (EoE) presents with symptoms of esophageal dysfunction and is characterized by large numbers of intraepithelial eosinophils in esophageal biopsies (1). Although it constitutes an increasingly reported cause for chronic dysphagia and food impaction, EoE remains unrecognized in many settings. In fact, some EoE patients have been diagnosed only after suffering non-traumatic esophageal perforation or Boerhaave's syndrome (2).

A 46-year-old male who had presented an episode of meat impaction resolved by endoscopic removal 5 years before was admitted to the emergency room after suffering a new food impaction episode. The endoscopy revealed a narrow, trachealized esophagus with a piece of meat in the lower third and a deep mucosal tear. After presenting sudden retrosternal pain, crepitus was noted upon palpation of the cervicothoracic region. A transhiatal esophagectomy was performed, reconstructing the distal esophagus by means of tubular gastroplasty.

Macroscopic study of the esophagetomy specimen showed thickened walls and stricture (**Figure 1a**, hematoxilin and eosin ×20). A lineal ulcer was observed in the proximal esophagus (**Figure 1b**, H&E ×2) at the site of impaction. Histological examination showed a dense neutrophilic infiltration and necrosis beneath the ulcer. Numerous samples were taken around the site of the ulcer and from the esophageal segment between the upper and lower thirds. Histological findings were compatible with EoE and consisted of diffuse eosinophilic infiltration of squamous mucosa (**Figure 1c**, H&E ×10) with a density >20 per HPF throughout the length of the esophagus, but predominantly in the medium and upper thirds. There were several microabscesses on luminal portions of the mucosa with reactive acanthosis of the epithelium and basal layer hyperplasia. Eosinophilic infiltration was observed throughout the esophageal wall, penetrating the submucosa (where lymph nodes were seen, **Figure 1d**, H&E ×10), dissecting muscle fibers of inner muscularis propria (**Figure 1e**, H&E ×4), and reaching parasympathetic ganglion cells of the myenteric plexus (**Figure 1f**, H&E ×40) (**Figure 1**).

Trichrome stains revealed extensive fibrosis of the subepithelial layer and hypertrophy of muscularis mucosae, both responsible for the increased wall thickness (**Figure 1g**, Masson trichrome ×4).

This case represents the first histopathological evaluation of the entire esophageal wall in a patient with EoE. Because endoscopic biopsy forceps usually sample above the lamina propria, the eosinophilic infiltration and fibrous remodeling of deep esophageal layers was only a supposition based on murine samples and extrapolated from the pathophysiology of bronchial asthma (3). We observed that eosinophils permeate the submucosal and muscle layers and the neuronal plexus, promoting fibrous remodeling, intense collagen deposition, and smooth muscle hypertrophy, altering the mechanical properties of the esophageal wall and reducing esophageal distensibility in EoE patients. Cytotoxic proteins in the eosinophil granules can lead to tissue damage, increasing the risk of esophageal disruption. These phenomena all have important clinical implications, as they determine dysphagia and explain not only strictures and dysmotility, but also the deep esophageal tears and perforation, resulting from vomiting to dislodge impacted food as frequently described in EoE patients.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

REFERENCES

1. Liacouras CA, Furuta GT, Hirano I *et al.* Eosinophilic esophagitis: updated consensus recommendations for children and adults. J Allergy Clin Immunol 2011;128:3–20.
2. Lucendo AJ, Friginal-Ruiz AB, Rodríguez B. Boerhaave's syndrome as the primary manifestation of adult eosinophilic esophagitis. Two case reports and a review of the literature. Dis Esophagus 2011;24:E11–5.
3. Aceves SS, Broide DH. Airway fibrosis and angiogenesis due to eosinophil trafficking in chronic asthma. Curr Mol Med 2008;8:350–8.

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Steinstrasse Formation After Extracorporeal Shock Wave Lithotripsy for Pancreatic Stones

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To the Editor: A 47-year-old man was admitted to our hospital due to intermittent