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Management of Esophageal Stricture and Perforation Complicated by Undiagnosed Eosinophilic Esophagitis and Pill Impaction

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BACKGROUND

Eosinophilic esophagitis (EoE) is a chronic-immune-mediated disease, characterized by esophageal dysfunction secondary to eosinophilic infiltration. EoE is now the leading cause of dysphagia and food impaction in both children and young adults (1). The incidence and prevalence is rising, with an estimated pediatric incidence of 0.7 to 10 cases per 100 000 person-years (2). Genetic predisposition, gene-environment interactions, and comorbidities increases one's risk of developing disease (3).

EoE affects all ages, with clinical presentation varying with age (1). Infants may present with failure to thrive and food refusal, whereas adolescents typically present with dysphagia and impaction (1). Diagnosis can be challenging due to symptoms that can mimic or occur concomitantly with symptoms of other diagnoses or be unintentionally masked by adaptive techniques (1–3). Schoepfer et al report that younger patients have longer diagnostic delay, with a gradual decrease in delay with increased age. Under-recognition of disease can lead to a median delay of up to 6 years and increase one's risk of complications, including fibrostenosis and stricture development (4).

This case describes a perforation in an esophageal stricture secondary to undiagnosed EoE and subsequent successful management. We highlight the need for increased disease recognition, and the complications associated with delayed diagnosis.

CASE

A 14-year old female patient, with known seasonal and tree nut allergies, presented to the Emergency Department overnight with acute onset globus sensation after ibuprofen pill ingestion. She had unsuccessfully attempted to relieve the sensation by drinking water and inducing emesis. She had an unremarkable examination, but she was admitted due to oral fluid intolerance. Further assessment revealed a history of 2 similar events in the last year, following a pill and steak ingestion. On both occasions, emesis provided relief; therefore, medical attention was not sought.

On further questioning in the morning, she reported increased water volume consumption with meals, slow food ingestion, and a softer food preference, prompting an expedited work-up. A barium esophagram revealed complete esophageal obstruction, with no passage of contrast past 3 to 4 cm inferior to the thoracic cleft (Fig. 1A). Esophagogastroduodenoscopy revealed evidence of EoE with significant trachealization, esophageal structuring, and an ibuprofen pill impaction (Fig. 1B). Pill extraction revealed a pill with a partially worn away coating and a stricture extending the length of the esophagus (Fig. 1C), through which a 5-mm scope could not pass. Maloney dilators were used to progressively dilate the stricture to 7 mm in diameter, with moderate resistance. Once dilated, further evidence of EoE and normal gastric mucosa was seen. Following dilation, 2 mucosal tears distal to the previous impaction (2 and 7 cm in length) and an esophageal perforation at 25 cm (Fig. 2A) were also visualized. Fluoroscopy confirmed a contained leak, without evidence of a pneumomediastinum (Fig. 2B). A nasogastric (NG) tube was placed under direct endoscopic visualization. She remained hemodynamically stable and was treated conservatively; with a proton pump inhibition and a 5-day course of ampicillin/sulbactam. She was placed nil per os with intravenous fluids initially, before transitioning to an elemental milk-free formula bolus regimen via NG tube. Repeat esophagram 4 days postprocedure revealed a 6-mm contained linear leak to the left of the midthoracic esophagus (Fig. 2C). She was discharged home on a proton pump inhibition twice daily and NG feeds.

Repeat esophagram 3 weeks later showed a stable leak. At 2 months, when the leak had resolved, Budesonide twice daily was started and her diet was slowly advanced from clear liquid to soft diet (remaining dairy-free throughout), with NG tube removal. Esophagogastroduodenoscopy at 4 months revealed EoE remission; significantly improved trachealization and impressive mucosal healing from her stricture dilation and subsequent complications. A small diverticulum with an epithelized base was present at the site of her prior leak (Fig. 3A) and a healed scar from the dilation-induced mucosal tear at 30 cm (Fig. 3B). At 4 and 10 months, respectively, a 9.2- and 9.9-mm scope were successfully passed, without need for dilation. She continues to have no limitation to bite size or consistency, with resolution of dysphagia symptoms.

DISCUSSION

Esophageal stricture is a serious EoE-related complication, with a varying reported incidence in pediatric patients; 0.2% to 28% (5). Strictures can be managed medically, but mechanical dilation should be considered as an appropriate therapeutic addition in those with severe dysphagia symptoms and to avoid further complications, including impaction (6). Additional studies are required to determine the preferred timing of dilation; at the time of diagnosis versus following medical treatment initiation, but the reported risk of adverse effects is comparable (6). Pediatric data on dilation-related perforations in patients with EoE is limited with no reported incidence. Nonetheless, the risk in those with and without EoE is comparable,

Received January 19, 2023; accepted August 1, 2023.
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The authors report no conflicts of interest.

JPGN Reports (2023) 4:4(e360)
ISSN: 2691-171X
DOI: 10.1097/PG9.0000000000000360

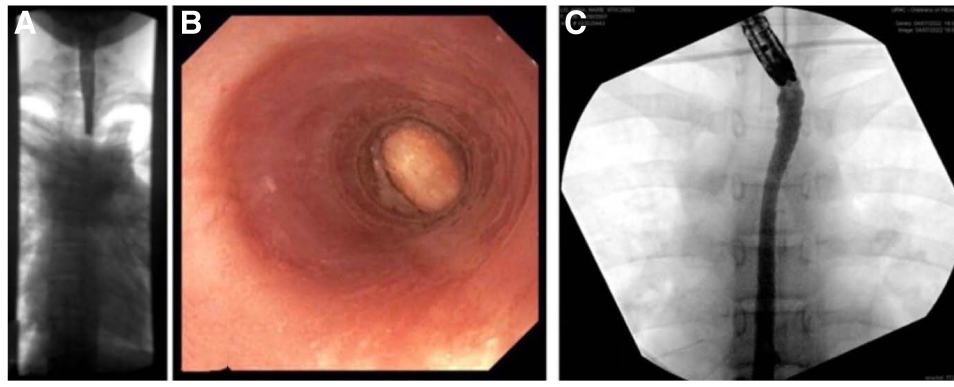

FIGURE 1. A) Barium esophagram showing complete esophageal obstruction. B) EGD showing significant trachealization, esophageal structuring, and an ibuprofen pill impaction. C) Barium esophagram showing stricture extending the length of the esophagus postpill extraction. EGD = esophagogastroduodenoscopy.

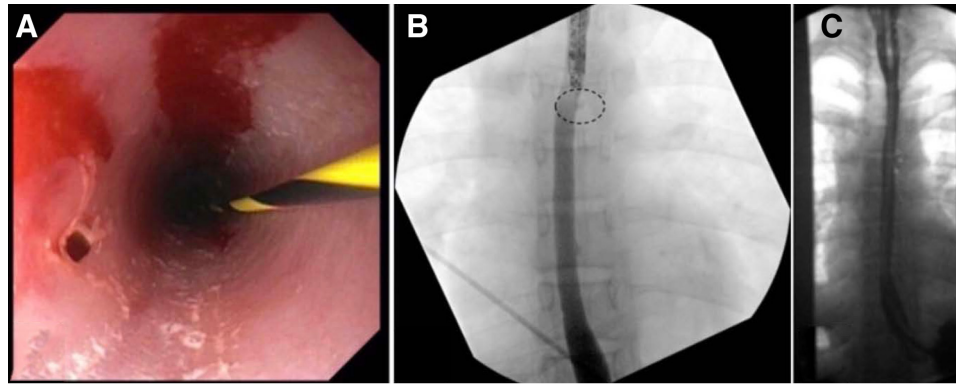

FIGURE 2. A) EGD showing an esophageal perforation postesophageal dilation. B) Fluoroscopy confirming a contained leak, without evidence of a pneumomediastinum. C) Repeat esophagram 4 days postprocedure revealing a contained leak. EGD = esophagogastroduodenoscopy.

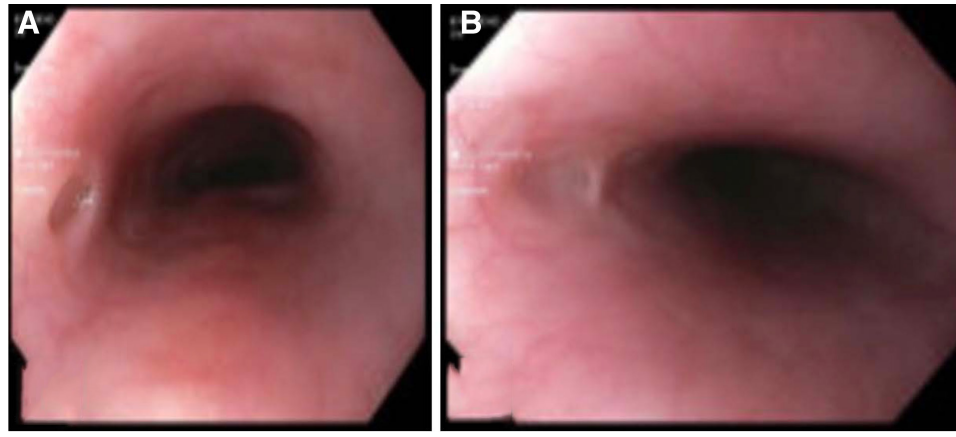

FIGURE 3. A) Healed mucosa; small diverticulum with an epithelized base at the site of her prior leak. B) Healed mucosa; scar from the dilation-induced mucosal tear.

and the risk of adverse events, including perforation, is considered uncommon but possible (6).

The duration of diagnostic delay directly correlates with stricture prevalence at the time of EoE diagnosis, with each year of symptoms before diagnosis increasing the risk of fibrostenosis by up to 5% (3,4). The risk of esophageal perforation in the setting of an EoE-related stricture is uncommon. However, this patient may have been at higher risk of perforation due to other factors including prolonged impaction and exposure to ibuprofen, which may have caused local inflammation, secondary to under-recognition of EoE on admission (7). Although a conservative approach may have avoided iatrogenic perforation, given her severe stenosis, symptoms, and the overall rarity of perforation, dilation at the time of pill removal was deemed appropriate.

Additionally, conservative management of the perforation and aggressive treatment of EoE ultimately led to esophageal healing and resolution of dysphagia.

Increased understanding of this disease and the development of clinical practice guidelines in Europe has resulted in decreased EoE diagnostic delay (8). This has positively impacted on the severity of symptoms and endoscopic features at diagnosis and the number of endoscopies needed to reach diagnosis (8). Nonetheless, there is still room for improvement.

Awareness of at-risk individuals and the symptoms of EoE is crucial for making an earlier diagnosis. Interrogating eating habits could prompt earlier investigation, including esophagram to rule-out impaction in an acute setting, and determine the need for urgent intervention.

Ultimately, earlier diagnosis would likely decrease the risk of complications.

ACKNOWLEDGMENTS

Consent was obtained from the father of the patient.

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