

Long-Lasting Effect of Endoscopic Dilatation of an Esophageal Stenosis due to Eosinophilic Esophagitis

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Abstract

Eosinophilic esophagitis is a rare disorder mainly affecting pediatric patients, although the number of cases reported in adults, especially young males with dysphagia, is on the increase. The most severe complication is esophageal stenosis. We report the case of a 26 year old white male who presented with dysphagia in 2001. Endoscopy revealed an esophageal stenosis 35 cm aboral without signs of mucosal inflammation, that was dilated once. The patient was asymptomatic until 2004, when he presented again with dysphagia. Eosinophilic esophagitis was diagnosed. Dilatation was repeated. The further clinical course was uneventful with no more episodes of dysphagia.

Key words

Eosinophilic esophagitis - dysphagia - esophageal stenosis - dilatation

Rezumat

Esofagita eozinofilică este o entitate rară ce afectează predominant pacienții pediatrici, deși numărul de cazuri raportate în rândul adulților, în special bărbați tineri acuzând disfagie, este în creștere. Complicația cea mai severă este stenoza esofagiană.

Prezentăm cazul unui tânăr în vârstă de 26 de ani care s-a internat în serviciul nostru pentru disfagie instalată acut. Gastroscopia a revelat o stenoză situată la 35 cm aboral, fără semne de inflamație și s-a efectuat dilatarea endoscopică. Pacientul a fost asimptomatic până în 2004, când s-a prezentat din nou cu disfagie. De această dată s-a diagnosticat

esofagita eozinofilică și s-a repetat procedura de dilatare endoscopică. Evoluția ulterioară a fost favorabilă, fără alte episoade de disfagie.

Introduction

Eosinophilic esophagitis is a rare entity characterized by a dense eosinophilic infiltration of the esophageal mucosa with more than 15 eosinophils per high-power field on esophageal biopsies (1). Although already mentioned by Dobbins in 1977 as the esophageal manifestation of eosinophilic gastroenteritis (2), it was Attwood who first described eosinophilic esophagitis as an entity with distinct clinicopathologic features (3). Mainly affecting young males and paediatric patients with food allergies, its clinical presentation includes most frequently dysphagia (93%), food impaction (62%) and heartburn (23.6%) (1). We describe a patient with an esophageal stenosis due to eosinophilic esophagitis, that could be dilated with an unusual long-lasting effect.

Case report

A 26 year old white male presented with dysphagia in November 2001. He complained about this inability to swallow solid food. Regarding his personal pathologic history, he had no food allergies, chemical irritations or episodes of dysphagia in childhood, but several food impactions in 2001. Physical examination showed a healthy young man with a body mass index of 20 kg/m². All laboratory findings were in the normal range. Upper gastrointestinal endoscopy revealed an esophageal stenosis 35 cm aboral without signs of mucosal inflammation. Radiography demonstrated a stenosis of the last 10 cm of the esophagus with a praestenotic dilatation. CT revealed no tumor impression. Further evaluation with pH-metry showed no acid reflux. The manometric study described a non-specific motility disorder of severe

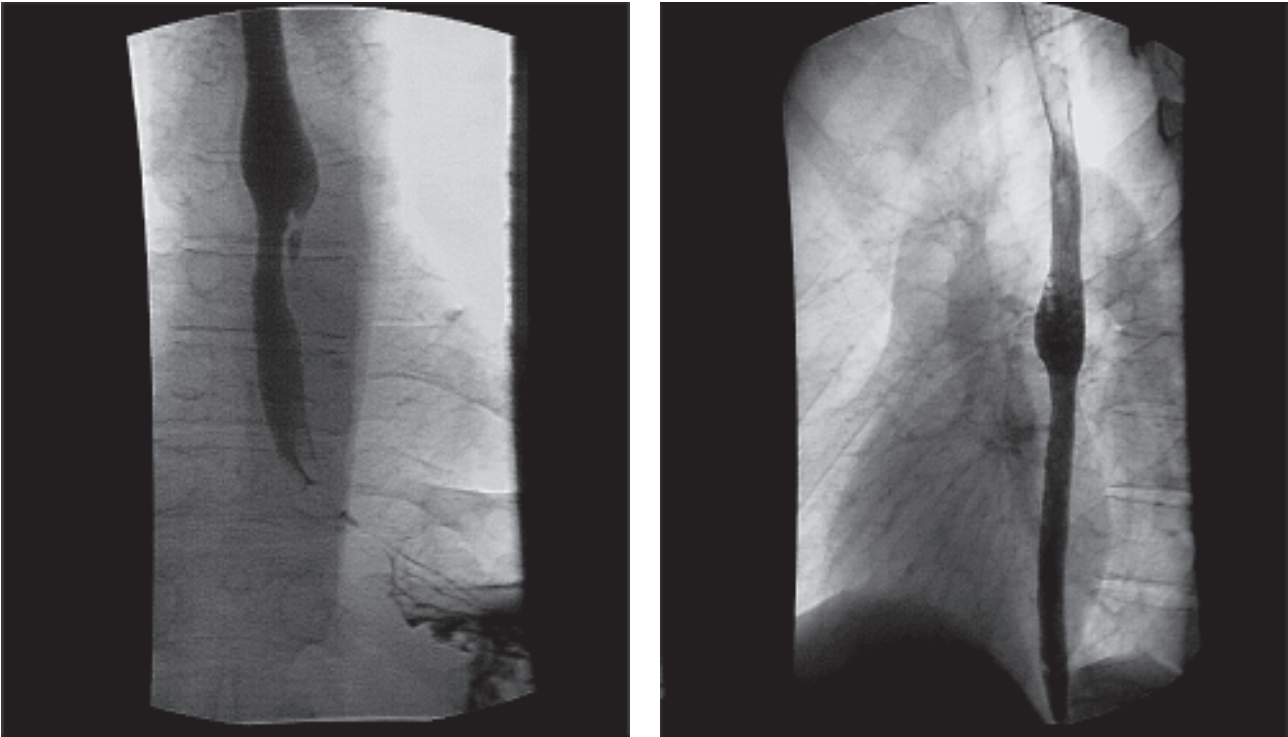


Fig.1 Esophageal stenosis with prestenotic dilatation before and after dilatation (2001).

intensity with abolished peristalsis compatible with the diffuse esophageal spasm or achalasia. The esophageal stenosis remained unexplained and was dilated once 11/01 (Fig.1). The patient was asymptomatic until 2004, when he presented again with dysphagia. This time a more extensive biopsy protocol was fulfilled. Considering the patient's age and gender, the esophageal stenosis without any signs of inflammation, the absence of reflux and the typical "red stripes" we suspected an eosinophilic esophagitis. Histology confirmed our clinical diagnosis. Because of the severe symptoms, dilation was performed before the histological diagnosis was established. After dilation the further clinical course was uneventful with no more episodes of dysphagia. Considering this fact as well as the long symptom free period after the first procedure, corticosteroid therapy seemed unnecessary. The patient presented only once again in the emergency room with an allergic reaction to nuts. The consultation of the allergologist for identifying further food allergens was recommended, but no results have been received yet.

Discussion

Eosinophilic esophagitis is an increasingly reported disease of both children and young males and is often associated with allergic reactions (2-5). It is controversially discussed if the allergic manifestations are IgE-mediated, "mixed" (involving some IgE- and some T-cell-mediated components) (8,9), or non-IgE-mediated (10). Spergel et al for example found that 73% of the patients with eosinophilic esophagitis had positive skin-prick tests, suggesting an

IgE-mediated mechanism, but also demonstrated by positive patch-tests in negative skin-prick tests patients a Th1-cell-mediated reaction (11,12).

The clinical presentation forms of the disease are numerous, ranging from dysphagia (most frequently found), food impactions, heartburn to nausea, vomiting, retrosternal or abdominal pain and even weight loss or cachexia. The symptoms are often misinterpreted as reflux disease. A typical clinical sign is the failure of acid-suppressing medication. Another distinct feature is the absence of acid reflux or episodic alkalinisations of the esophagus on pH-metry (13).

The manometric findings in eosinophilic esophagitis reveal motility disorders in 40% of the cases (1). These include non-specific motor abnormalities of severe intensity affecting the esophageal body, simultaneous waves and secondary peristaltic waves in three thirds of the organ (14). Other studies mention the existence of tertiary contractions, aperistalsis or diffuse esophageal spasms mimicking the "nutcracker" esophagus (15). An association between eosinophilic esophagitis and achalasia has also been described (16).

The endoscopic descriptions vary from a normal appearance (1,17,18) or non-specific abnormalities like mucosal oedema (1), granular mucosa, absence of the vascular design to more specific lesions such as red stripes, whitish pinpoint exudates or an extreme fragility of the mucosa, "crepe paper esophagus", considered by some authors as pathognomonic (19). Also regarded as pathognomonic for eosinophilic esophagitis is the presence of strictures which contain multiple, closely spaced, ring-like indentations, inducing the appearance of esophageal

“trachealisation”, the so-called “ringed esophagus” (7,14,20,21). Other authors also describe the presence of regular stenosis (14,21,22), whether focal (23) or extensive, giving the aspect of “small caliber esophagus” (24).

The entity can only be diagnosed by multiple biopsies. Although the typical histological finding in eosinophilic esophagitis is the dense infiltration of the mucosa with at least 15 eosinophils per high-power field (1), many authors considering a number of > 20 eosinophils/hpf as necessary for diagnosis (17,18,21). The presence of eosinophilic microabscesses seems to be characteristic (17,25), corresponding to the popular whitish exudates seen on endoscopy (17). Obviously fibrosis must also be part of the disease, leading to stenosis.

Regarding the treatment, most of the authors recommend, based on the pathophysiological mechanism, dietary measures as first option. These range from complete dietary elimination and use of L- amino acid-based formula or protein hydrolysates to identified food allergens elimination (8,9,12,18,26). When all dietary measures are unsuccessful, corticosteroids are indicated as second-line treatment (8). Usually, topical steroids such as fluticasone propionate are used with good results and no adverse effects (8,14,26,27). As maintenance therapy in patients with known allergies, dietary restriction is most effective, but montelukast and topical cromolyn may also be useful (26). Systemic corticosteroids are reserved for more severe cases (8, 26). In these situation, symptomatic improvement can also be achieved by endoscopic dilation procedures. The relief of dysphagia is only temporary (21), lasting normally about 7 months (28). As a recent review concluded, the bougienage has also an increased complication rate, which could be reduced by the previous use of corticosteroids (1).

The long-lasting effect of the dilatation in our case – three years – is a rarity, maybe due to a minor fibrosis.

Conclusion

Treatment of esophageal stenosis due to eosinophilic esophagitis with dilatation should be performed in patients with dysphagia due to severe stenosis, especially when the histological diagnosis has not been established or other therapeutic options failed. Despite the general opinion considering dilatation as a procedure with limited efficacy and high complication rate in eosinophilic esophagitis, in our case bougienage was safe, effective and has led to a long symptom free interval.

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