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Digestive and Liver Disease

Digestive and Liver Disease 37 (2005) 129-134

www.elsevier.com/locate/dld

Brief Clinical Observation

Ringed oesophagus and idiopathic eosinophilic oesophagitis in adults: an association in two cases

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Received 10 January 2004; accepted 8 April 2004 Available online 23 November 2004

Abstract

Ringed oesophagus is an increasingly recognised finding in young people presenting with dysphagia and may be related to eosinophilic oesophagitis. Recently, hypotheses regarding potential aetiologies have been proposed but these have not been systemically tested in the majority of reported cases. We report two cases very similar in clinical history and endoscopic findings. An association with gastro-oesophageal reflux disease or motility abnormalities of the oesophagus were ruled out in both. Histological analysis revealed high-density infiltration of the oesophageal mucosa by eosinophils and excluded gastro-duodenal involvement. Examinations of the oesophagus at the time of low frequency dysphagia, some years before presentation to our centre, did not show rings, suggesting that multiple rings are a possible late complication of eosinophilic oesophagitis. Oesophageal dilatation effectively relieved dysphagia in our two patients.

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Keywords: Eosinophilic oesophagitis; Ringed oesophagus

1. Introduction

Multiple oesophageal rings, also known as ringed oesophagus, is an uncommon finding during upper endoscopy and is mainly detected in children [1] and younger adults [2,3]. Episodic dysphagia is the presenting symptom in most cases. The aetiology remains unknown in several cases and the association with gastro-oesophageal reflux disease and motility abnormalities of the oesophagus is controversial and not tested in the majority of the patients reported [4,5]. A congenital form of this disease has also been suggested [2,6]. Recently an association with eosinophilic oesophagitis in adults has been reported in some cases [7]. Therapy is based on dilatations of the oesophageal rings [2,3,8], corticosteroids [7] and immunomodulation [9]. We report two cases of multiple oesophageal rings in

which the association with gastro-oesophageal reflux disease and motility abnormalities was ruled out and possible causality by concomitant eosinophilic oesophagitis is discussed.

2. Case 1

A 36-year-old man presented to our centre complaining of recurrent episodes of dysphagia with solid food. These episodes began 10 years prior to presentation at our centre and occurred on a monthly basis. Neither heartburn nor atypical symptoms of gastro-oesophageal reflux disease were associated. No previous diseases, history of allergies or administration of drugs were reported. At the time of initial presentation with dysphagia he underwent an upper endoscopy that revealed a linear erosion at 22 cm from the mouth, while the mucosa at the oesophago-gastric junction was normal. An empirical course of PPI had no beneficial ef-

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Fig. 1. Barium X-ray revealed a narrowed middle third of the oesophagus with a modest proximal dilatation, where multiple rings (arrows) were suspected.

fect. Since then the dysphagia slowly worsened in frequency becoming 3–4 episodes/week at the time of evaluation in our centre. Body weight was stable (body mass index 21). A barium X-ray revealed a narrowed oesophagus in its middle third with a modest proximal dilatation (Fig. 1); at the level of narrowing the radiologist suspected the presence of multiple rings. An oesophageal manometry definitively ruled out achalasia but revealed the absence of motor activity between 18 and 10 cm above the lower oesophageal sphincter (LOS), a level corresponding to the narrowing (Fig. 2). A 24-h pH measurement was normal: percentage of time at pH < 4 was 1.5 (n.v. in our laboratory: <5.5). Upper endoscopy confirmed multiple rings at the middle third of the oesophagus, surrounded by normal-appearing mucosa (Fig. 3), with a modest resistance to the passage of the scope (GIF145; Olympus

Optical Co., Hamburg, Germany). Multiple biopsies were obtained at the level of the multiple rings, the caudal oesophagus, stomach and second portion of the duodenum. During the same endoscopic session an oesophageal dilatation was performed with Celestin dilators, up to 18 mm. Passage of the endoscope after dilatation revealed disappearance of the rings and multiple longitudinal mucosal tears (Fig. 4). After 6 h of monitoring the patient was discharged without complications. The histopathological analysis revealed high-density eosinophilic infiltration (30 eosinophils per high-powerfield) of the oesophageal mucosa at all levels (Fig. 5). No submucosal layer was present. No eosinophilic infiltration was detected in the stomach and duodenum and the diagnosis was ringed oesophagus associated with eosinophilic oesophagitis. A manometry was performed after dilatation and

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