CASE REPORT

Oesophageal lichen planus

Paulo Souto[1], Carlos Sofia[1], José Pina Cabral[1], António Castanheira[1], Sandra Saraiva[1], Oscar Tellechea[2], António Donato[1] and Diniz Freitas[1]

Lichen planus is a common skin and mucosal disease, with very rare symptomatic oesophageal involvement. We report a case of painful dysphagia due to oesophageal lichen planus in a 60-year-old woman who also had oral, cutaneous and genital lichen planus lesions. Steroid treatment produced considerable improvement of all lesions and a rapid symptomatic remission.

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Introduction

Lichen planus is a common inflammatory mucocutaneous disorder of unknown aetiology, with frequent oral involvement occurring in 30–70% of patients [1]. With few cases reported, oesophageal disease may result in painful dysphagia or stenosis.

Case report

A 60-year-old woman, with a 4-year history of dysphagia and odynophagia, especially for solids, was admitted to the gastroenterology department. There was no history of heartburn or regurgitation. Despite treatment with ranitidine and sucralfate, no symptomatic benefit occurred and endoscopic upper oesophageal lesions described as erosive oesophagitis persisted. At the onset of her oesophageal symptoms, she began to complain of vulvar pruritus and mouth pain, and she also noticed exacerbation of old skin lesions of the lower back. There was a family history of pulmonary tuberculosis.

Physical examination revealed erosive lesions of the oral mucosa and an ulcerative vulvovaginitis. On the lower back, erythematous and violaceous papular skin lesions were present together with areas of hyperpigmentation (Fig. 1). Laboratory studies and chest x-ray were normal. A barium swallow revealed an irregular mucosal surface and poor distensibility of the proximal oesophagus. At endoscopy, there were severe erosive changes extending to 26 cm from the incisors (Fig. 2). In the lower oesophagus, hyperaemic and friable areas were present, covered with a white membranous layer easily stripped with minimal

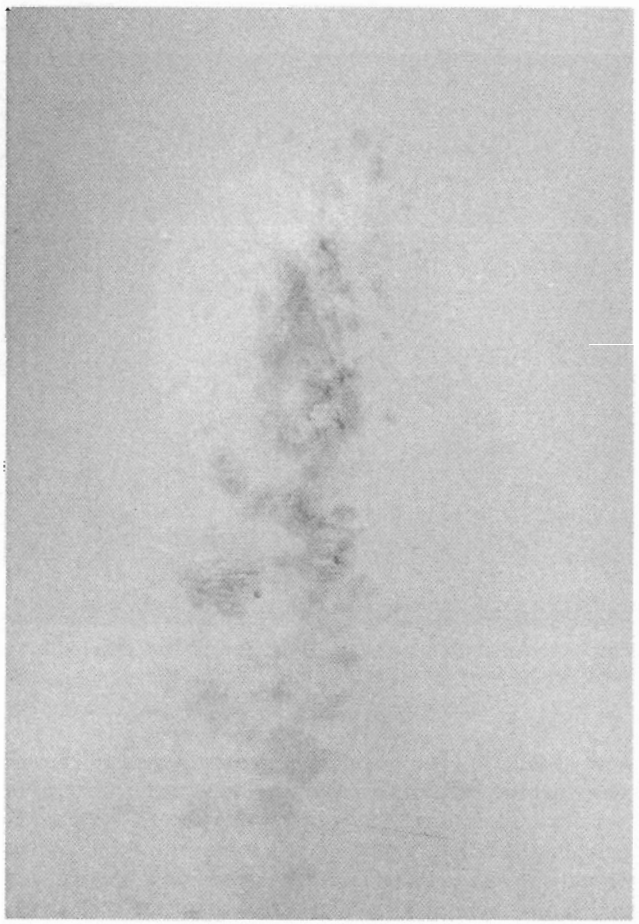

Fig. 1. Erythematous and violaceous papular skin lesions and areas of hyperpigmentation on the lower back.

From the Departments of [1]Gastroenterology and [2]Dermatology, Coimbra University Hospital, Coimbra, Portugal.

Requests for reprints to Paulo Souto MD, Department of Gastroenterology, Coimbra University Hospital, 3049 Coimbra Codex, Portugal.

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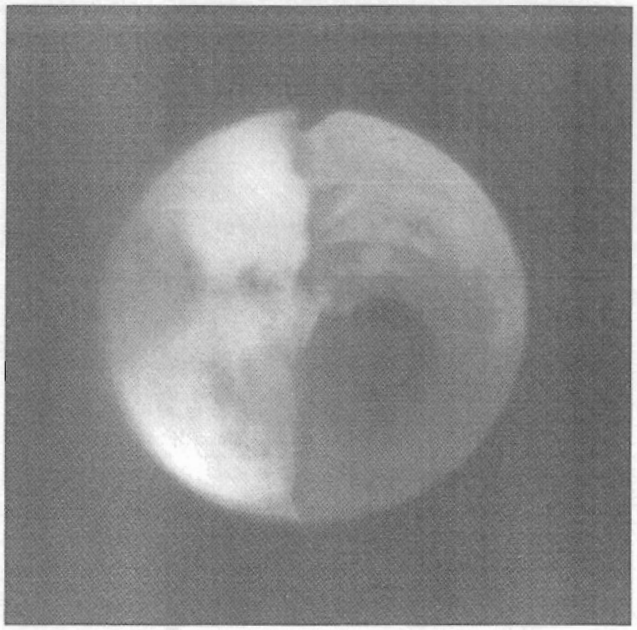

Fig. 2. Severe erosive lesions in the proximal oesophagus.

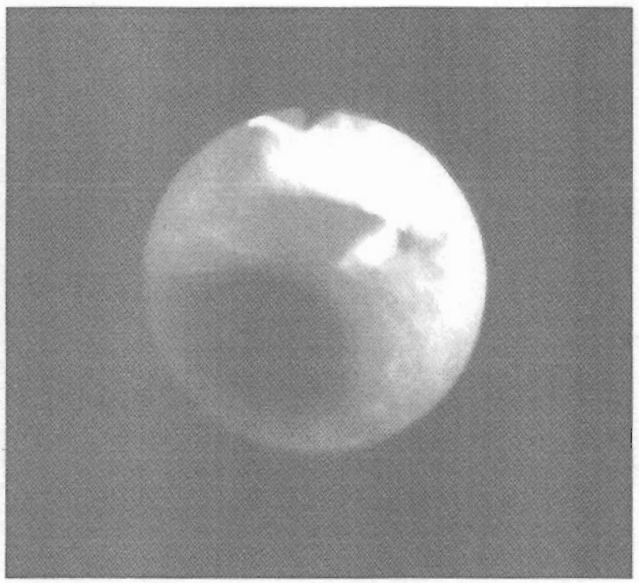

Fig. 3. Hyperaemic area in the distal oesophagus covered by a membranous layer clearly stripped.

contact (Fig. 3). The biopsies were first reported as 'chronic non-specific inflammatory changes, without criteria of peptic oesophagitis'. Scintigraphy showed no gastro-oesophageal reflux. Computed tomography of the mediastinum showed unremarkable thoracic organs. Oesophageal endoscopic ultrasonography revealed no changes. A tuberculin intradermal test was done which produced an exuberant bullous reaction. Attempts to detect acid-fast rods in gastric aspirate, oesophageal biopsies or bronchoalveolar washings were negative. There was no response to antituberculosis therapy. Tissue culture for fungal infection was also negative. Biopsies of the vaginal mucosa revealed an ulcerated chronic inflammation. The macroscopic and histological appearances of the skin lesions were consistent with lichen planus. A second look examination of oesophageal biopsies revealed an extensive band-like lymphocytic infiltrate, consistent with lichen planus.

Systemic retinoid therapy, 50 mg etretinate per day, orally, was initiated but stopped 1 month later because of side effects and the lack of any apparent symptomatic or endoscopic benefit. Treatment with oral methylprednisolone produced rapid symptomatic improvement, with disappearance of dysphagia. Any attempt to withdraw steroids resulted in recurrence of dysphagia and exacerbation of all the lichen planus lesions. At 3-year follow-up, she is taking 6 mg deflazacort per day. In repeated endoscopic examinations, even with higher doses of steroids, discrete papular lesions are still present. Periodic cytology samples are taken, with no evidence of malignancy.

Discussion

Oesophageal symptomatic lichen planus is a rare condition [2]. With few cases described since the first reports in 1982 [3,4], the prevalence of oesophageal disease was underestimated until an endoscopic study revealed oesophageal involvement, usually subtle, in a quarter of patients with lichen planus [1]. All the cases were described in women presenting with dysphagia or odynophagia [2]. Usually affecting the proximal oesophagus, it may result in the formation of benign strictures [5]. The endoscopic lesions are described as erosions or friable haemorrhagic mucosa covered by an easily stripped membranous layer [6], both features being present in our case (Figs 1 and 2). Oesophageal involvement may precede other lesions [7], making the diagnosis even more difficult. The histological picture is quite different from reflux or fungal oesophagitis [1,6]. In spite of its rarity [8], in the presence of an exuberant reaction to the tuberculin skin test and a family history of tuberculosis, a tuberculous aetiology should be excluded. Acid-fast rods were not detected in our patient and no response occurred with antituberculosis therapy.

Although there are no reported cases of malignant change in oesophageal lichen planus, follow-up is advised [5,9], considering the malignant potential of oral lichen planus [10,11]. Steroid treatment may reduce this potential risk and provides rapid symptomatic improvement [7,12]. A variable response is achieved with retinoids [11,13,14].

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