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Clinical Record

Oesophagitis dissecans superficialis complicating repeated rigid oesophagoscopy and dilatation

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Abstract

Oesophagitis dissecans superficialis is an extremely rare and benign condition where the mucosal epithelium of the oesophagus sloughs off along the whole length of the oesophagus and is expelled as an oesophageal cast. This condition has been reported in association with various aetiological factors. We report a case of an oesophageal cast in a patient who underwent repeated oesophagoscopy and dilatation for a postcricoid web. We discuss the possible relationship between trauma to the upper oesophagus and the development of oesophagitis dissecans superficialis.

Key words: Oesophagoscopy; Oesophagitis dissecans superficialis; Oesophageal cast, postcricoid web

Introduction

Oesophagitis dissecans superficialis, also known as oesophageal cast, is an entity where a part or the entire superficial mucosal lining of the oesophagus sheds away. The epithelial layer of the oesophageal mucosa separates at the epithelium-lamina propria junction,[1] sloughs off and is expelled as a long thin membranous tube. This oesophageal mucosal loss with resultant sloughing of an intact epithelial cast is extremely rare. The condition was first described by Birsch-Hirschfeld in 1877 and was subsequently named 'oesophagitis dissecans superficialis' by Rosenberg in 1892.[2] Oesophageal cast phenomenon has been observed in infants as well as in elderly patients. The condition is thought to be secondary to trauma (chemical, thermal or physical) and different dermatological diseases including exfoliative dermatitis, angina bullosa haemorrhagica, bullous pemphigoid, epidermolysis bullosa acquisita, pemphigus vulgaris, severe generalised eczema[3] and Graft-versus-Host disease. We report a case of oesophageal cast as a result of iatrogenic trauma from repeated rigid oesophagoscopy for dilatation of a postcricoid web.

Case Report

A 62-year-old lady presented with dysphagia to solid foods of two months' duration. There was no history of loss of appetite, weight loss, gastroesophageal reflux or dermatological problems. All the routine laboratory tests were within normal limits. A barium swallow confirmed the presence of a postcricoid web. She underwent repeated rigid oesophagoscopies and dilatation of the postcricoid web with gum elastic bougies almost on a yearly basis. On the ninth oesophagoscopy, polypoidal granulation tissue was found immediately below the cricopharyngeal sphincter. Whilst trying to obtain a biopsy of the granulation tissue, a 30 cm long thin walled collapsed tube of oesophageal mucous membrane came out (Figure 1). The membrane remained attached at one point to the cricopharynx and had to be cut off. No oesophageal bleeding was noted following removal of the cast. Microscopy of the specimen showed non-keratinized squamous epithelium consistent with an oesophageal cast. (Figure 2). There was no evidence of mucosal disease, atypia or inflammation. A nasogastric tube was then kept in place for 48 hours. The patient made an uneventful post-operative recovery and was discharged home. A repeat oesophagoscopy after three months showed regenerated normal oesophageal mucosa.

Discussion

To date there have been 39 cases of oesophageal cast reported in world literature. The aetiological factors implicated include trauma to the oesophagus due to alcohol abuse, swallowing hot drinks, spicy foods, swallowing large quantities of food quickly,[3] ingestion of caustic agents, repeated forceful bouts of vomiting, Mallory–Weiss syndrome, nasogastric intubation[4] and oesophageal sclerotherapy.[1] In all these cases it is speculated that the separation of the cast is gradual as it detaches from the distal to the proximal oesophagus with healing occurring very quickly.

Oesophagitis dissecans superficialis is also associated with dermatological conditions like pemphigus vulgaris,[3,5] pemphigoid,[6] and epidermolysis bullosa acquisita.[7] Associations with oesophageal sclerotherapy,[1] oesophageal strictures,[8] alendronate therapy and graft versus host reaction[9] have been documented.

Patterson[10] described a simple oesophageal cast without an obvious aetiology and collected 17 other reported cases. To explain the mechanism of formation of an oesophageal cast, he suggested that initial trauma, thought probably due to dietetic indiscretions, sets up a low grade, chronic, superficial oesophagitis which caused an excessive epithelial proliferation and underlying oedema, followed by a certain amount of degeneration in the epidermoid layer.

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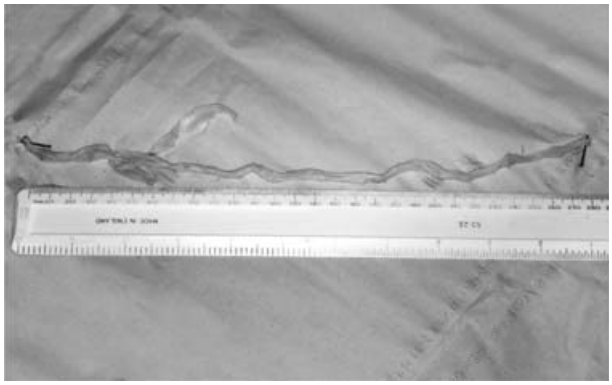

FIG. 1
Gross photograph of the oesophageal cast.

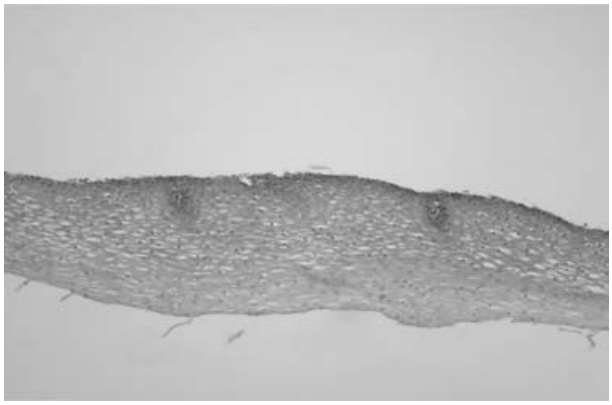

FIG. 2
Photomicrograph of oesophageal cast showing non keratinised squamous epithelial lining (H&E; ×10).

At the constriction near the distal end of the oesophagus, a process of slow annular ulceration sets in, which subsequently breaks the continuity of the mucosa at this level. Elsewhere there is a variable degree of milder inflammatory reaction with patchy exudation of serum into the sub-epithelial layer, serving to loosen the epithelium. Thereafter, periodical contraction of the muscular coat further aggravates the condition, detaching the lining in many places. Progressive mechanical exfoliation then goes on from below upwards throughout the whole length of the gullet. According to Patterson, the cast separated from the distal end because the trauma site was the distal oesophagus.

- **Oesophagitis dissecans superficialis is an extremely rare and benign condition where the mucosal epithelium of the oesophagus sloughs off along the whole length of the oesophagus and is expelled as an oesophageal cast**
- **This paper reports a case of an oesophageal cast in a patient who underwent repeated oesophagoscopy and dilatation for a postcricoid web**

Our case is unique, in that the trauma incurred was at the proximal end of the oesophagus rather than at the distal end. This trauma was caused by the rigid oesophagoscope and the dilators. We believe that the separation of the epithelium must have started at the proximal end and spread distally, resulting in the entire epithelial layer gradually stripping off along the whole length of the oesophagus. Our patient has been regularly followed up and we have noted that her symptoms of dysphagia improved after the cast was expelled. Interestingly, her symptom free period after the cast came out was significantly longer than that between the previous oesophagoscopies. It is known that the histology of a postcricoid web is essentially that of a fold of normal oesophageal epithelium with some underlying loose connective tissue.[11] We contemplate that the web had become less prominent after its epithelial layer sloughed off along with the oesophageal cast.

Conclusion

To our knowledge, there are no previous reports of oesophageal casts associated with postcricoid webs and repeated rigid oesophagoscopies. Awareness of the possibility of forming an oesophageal cast as a result of trauma due to repeated rigid oesophagoscopies and dilatations in the management of postcricoid webs is important.

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