Case reports

Lymphangioma of the oesophagus: a case report and review of the literature

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Abstract

Lymphangioma of the oesophagus is exceedingly rare. Seven cases (including our present case) have been reported in the world and are reviewed.

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Benign, submucosal tumours of the oesophagus, which are uncommon, may be derived from any of the structures in the oesophageal wall. Most of these tumours are leiomyomas. Vascular tumours account for only 2% of all the benign lesions of the oesophagus. Most of these are haemangiomas, while lymphangiomas are exceedingly rare. A definitive diagnosis of lymphangioma preoperatively is difficult to make and most are diagnosed only after surgical or endoscopic excision. A case of lymphangioma of the oesophagus is discussed with particular attention to indications for surgery.

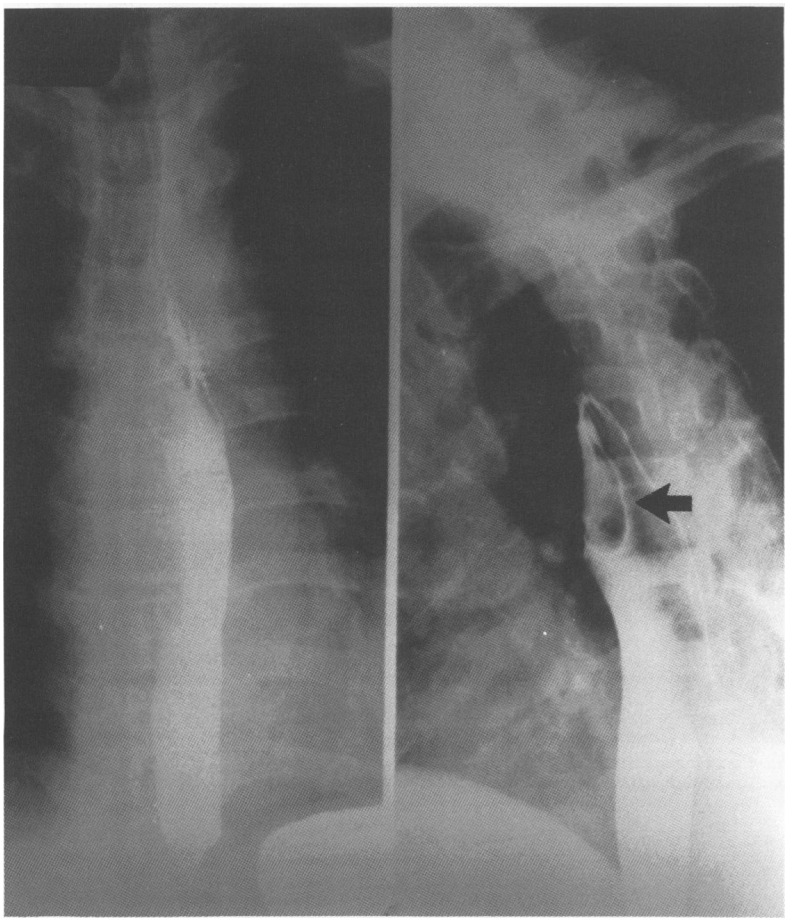

Figure 1 Oesophagogram of a patient with a lymphangioma showing a raised lesion in the middle third of the oesophagus, and a tiny dimpling of the covering mucosa (arrow).

Case report

A 55 year old man presented in 1992 with occasional, slight, retrosternal burning unrelated to swallowing. An upper gastrointestinal radiographic series revealed a raised lesion in the middle third of the oesophagus with no stenosis or irregularity of mucosal folds (fig 1). Endoscopy showed a submucosal mass on the anterolateral wall of the oesophagus, 25 cm from the incisors. No erosion, redness, or capillary dilatation were seen on the overlying mucosa. A biopsy was performed which was consistent with chronic oesophagitis without evidence of malignancy. Although a submucosal tumour, probably a leiomyoma, was suspected, the appearance was suggestive of malignancy, as it was 4 cm in diameter with a tiny dimpling of the overlying mucosa. A right thoracotomy was performed through the fifth intercostal space. The mid portion of the oesophagus was mobilised, but the tumour was not visualised, nor palpable within the oesophageal wall. Intraoperative endoscopic examination was necessary to identify its location. The tumour was isolated easily from the muscle layer, but not from the mucosa, and was therefore removed with the overlying mucosa. Histological examination of the tissue on intraoperative frozen section and postoperative paraffin section showed a cavernous lymphangioma without evidence of malignancy (fig 2).

Postoperative upper gastrointestinal radiography performed two weeks after surgery revealed normal morphology and function of the oesophagus. The patient was discharged well three weeks after the operation.

Discussion

Benign tumours of the oesophagus are very rare. Leiomyomas derived from the smooth muscle of the oesophageal wall account for most of these tumours.[1,2] Oesophageal lymphangioma is extremely rare. It was first described by Schmidt *et al* in 1961,[3] and in 1973 Brady and Milligan reported the first well documented case.[4] Only seven cases, including the present one, have been reported (table).[5–8] This tumour can be recognised by its projection into the

Summary of all the reported cases of lymphangiomas of the oesophagus

Author	*Year*	*Age*	*Sex*	*Symptoms*	*Location*	*Size (cm)*	*Endoscopic biopsy*	*Treatment*
Schmidt *et al*[3]	1961			Post mortem study. Data of cases unknown				
Brady and Milligan[4]	1973	62	F	Epigastric pain	Lower	5·0	Lymphangioma (Eder-Hufford oesophagoscope)	Conservative therapy
Armengol-Miro *et al*[5]	1979	64	M	Epigastric pain	Lower	1·0	Unknown	Endoscopic polypectomy
Tamada *et al*[6]	1980	46	M	Dysphagia	Middle–lower	Unknown	Not performed	Surgical conservative therapy
Liebert *et al*[7]	1983	58	M	Dysphagia	Lower	1·5	Unknown	Endoscopic polypectomy
Castellanos *et al*[8]	1990	66	F	Chest pain, heartburn	Middle	2·0	Unknown	Surgical enucleation
Present case	1994	55	M	Slight heartburn	Middle	4·0	Chronic oesophagitis	Surgical extirpation

lumen, pale colour, overlying normal mucosa, cystic translucency, and its deformation under pressure with the endoscope. However, endoscopically it is difficult to differentiate an oesophageal leiomyoma from oesophageal varices, haemangioma, or other submucosal tumours.

Symptoms may vary depending on the location of the tumour and degree of obstruction. Our patient compained of slight, retrosternal burning without dysphagia. Of all the cases reported, lymphangiomas were symptomatic in only two,[6 7] the main symptoms being epigastric pain, chest pain, and heartburn, presumably caused by gastric or duodenal ulcers, hiatus hernias, or reflux oesophagitis.

Biopsy may be necessary for definitive diagnosis, but it may not be possible because normal surface epithelium covers the tumour and can lead to misleading information from endoscopic sampling. In our case the biopsy specimen revealed only chronic oesophagitis. Brady and Milligan have used the Eder-Hufford oesophagoscope to provide sufficient tissue to establish the diagnosis of lymphangioma.[4] Tamada *et al*[6] reported that a biopsy was not performed because the lesion resembled oesophageal varices.

The treatment of oesophageal lymphangioma may be conservative if a definitive histological diagnosis is obtained and it is asymptomatic. No cases of malignant transformation have been described. Two of the reported cases were treated with polypectomy at the time of diagnostic endoscopy as the tumours were small (1 cm and 1·5 cm).[5 7] However, when the tumour is large, symptomatic, or if a malignant lesion is suspected, resection should be performed. The tumour in the present case was 4 cm in length and too wide for endoscopic polypectomy. There was also the possibility of malignancy. At surgery it can be difficult to determine the extent of tumour through the oesophageal wall, because a lymphangioma is soft and compressible and pushes into the lumen. We successfully used an endoscope to locate the tumour during the operation. We recommend the combination of surgery with operative endoscopy when the lymphangioma is unsuitable for endoscopic polypectomy.

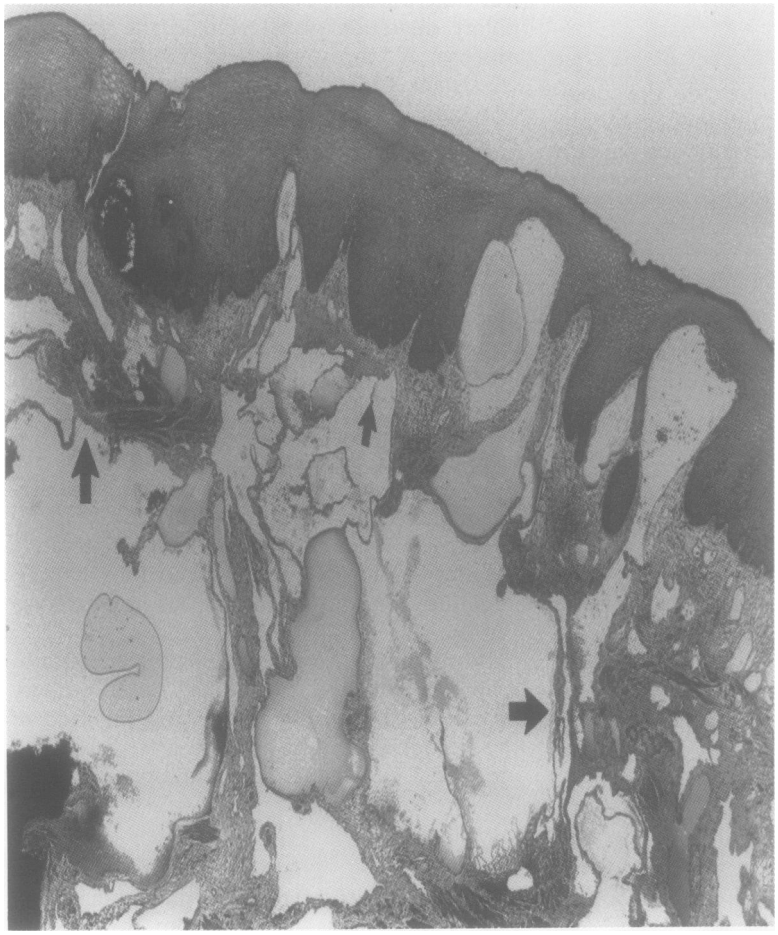

Figure 2 Photomicrograph showing normal oesophageal epithelium with proliferation of dilated lymphatic channels (arrows). Stain: haematoxylin and eosin; magnification × 12·5 reduced to 59% in origination.

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