CASE REPORT

Giant leiomyoma of the oesophagus with eosinophilic infiltration

Hadi Mutairi,¹ Mohammad Al-Akkad,¹ Mussarat Afzal,² Ikram Chaudhry¹

¹Department of Thoracic Surgery, King Fahad Specialist Hospital, Dammam, Saudi Arabia

²Department of Anaesthesia, King Fahad Specialist Hospital, Dammam, Saudi Arabia

Correspondence to
Ikram Chaudhry,
drihc@hotmail.co.uk

SUMMARY

The most common benign tumour of the oesophagus is leiomyoma. Haemopoietic elements rarely infiltrate oesophageal leiomyoma. We report the case of a 24-year-old man with a long history of intermittent dysphagia. Endoscopy revealed external compression with normal oesophageal mucosa. A barium swallow study showed a defect in the oesophageal wall and a narrow oesophageal lumen. A CT scan of the chest confirmed a mass in the mid-oesophagus. A tumour was excised and the histology report revealed leiomyoma infiltrated with eosinophils, which is a rare variant.

BACKGROUND

Leiomyomas are rare benign neoplasms of the oesophagus. They constitute less than 1% of oesophageal neoplasms and almost 10% of the mesenchymal tumours in the gastrointestinal tract.¹ Stromal and mesenchymal tumours of the digestive tract typically present as subepithelial tumours, which are broadly divided into two groups, the more common one being collectively called gastrointestinal stromal tumours. A less common group of mesenchymal gastrointestinal tract neoplasms consists of a spectrum of tumours that are identical to those arising in the soft tissue throughout the rest of the body. They include lipoma, leiomyoma, leiomyosarcoma, leiomyoblastoma, schwannoma and peripheral nerve tumours based on the histopathological findings.² Oesophageal tumour commonly presents as obstructive dysphagia, while the other causes of functional dysphagia like peptic stricture, achalasia and motility disorders, should be excluded. Diffuse oesophageal leiomyomatosis in association with idiopathic eosinophilic and allergic eosinophilic esophagitis has been reported previously.^{3–4} We report a rare case of a solitary giant oesophageal leiomyoma infiltrated by eosinophils.

CASE PRESENTATION

A 24-year-old man presented with a 2-year history of intermittent dysphagia which became worse 4 months prior to presentation. He had no history of chest pain, vomiting, weight loss, dyspepsia or oesophageal reflux disease. He also had no history of allergy, eczema or any other disease or surgery. Physical examination of the chest was normal. Basic blood investigations including a full blood count, liver and renal panels were normal. The absolute eosinophil count was $0.14 \times 10^9/L$ (normal range is $0.1–0.5 \times 10^9/L$).

INVESTIGATIONS

- Barium swallow showed a filling defect in the mid-oesophagus (figure 1A).
- CT scan of the thorax revealed a narrow oesophageal lumen with external compression (figure 1B).
- Endoscopy showed normal oesophageal and gastric mucosa and an oesophageal lumen narrowed by external compression at 21–29 cm of the oesophagus. A mucosal biopsy was not taken keeping in view that the endoscopic and radiological findings were highly suggestive of leiomyoma.

Endoscopic ultrasound is a very useful diagnostic tool in such cases, but it was not available.

DIFFERENTIAL DIAGNOSIS

- Leiomyoma
- Carcinoma of the oesophagus
- Achalasia
- Peptic stricture.

TREATMENT

The oesophagus was approached through a right posterolateral thoracotomy. On palpation, there was a large fusiform mass in the mid-oesophagus which displaced the nasogastric tube sideways. A longitudinal esophagotomy incision was made in the anterolateral oesophageal wall and the muscle layers were divided meticulously keeping the mucosa intact. An 8 cm mass was enucleated with careful dissection (figure 2A). The oesophageal wall was closed with '2–0' vicryl sutures. Postoperative recovery was uneventful. The histopathological report showed a well-circumscribed benign smooth muscle tumour heavily infiltrated with eosinophilic granulocytes (figure 2B). Immunohistochemical studies showed that the tumour cells were smooth muscle antigen (SMA) positive, CD 34 negative and CD 117 negative.

OUTCOME AND FOLLOW-UP

After excision of the oesophageal leiomyoma, the patient made an uneventful recovery. Histopathology showed a rare variant of oesophageal leiomyoma infiltrated with eosinophils. The follow-up barium swallow (figure 1C) and CT scan of the chest were normal (figure 1D).

DISCUSSION

Smooth muscle mesenchymal tumours of the oesophagus are classified as leiomyoma, leiomyosarcoma and leiomyoblastoma, based on the histopathological pattern, cellularity and mitotic

CrossMark

To cite: Mutairi H, Al-Akkad M, Afzal M, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-201343

Figure 1 (A) Barium swallow study preoperatively showing a mass arising from the wall of the oesophagus (arrow). (B) CT scan of the thorax showing a mass in the wall of the oesophagus displacing the lumen (arrow) with normal surrounding structures. (C) Postoperative barium swallow showing a normal oesophagus. (D) Postoperative CT scan of the thorax showing a normal oesophageal lumen.

activity.^{5 6} Leiomyoma is a very rare benign oesophageal tumour. Kimura *et al*⁷ reported six cases of oesophageal leiomyoma from 1973 to 1996. The peak incidence of leiomyoma is between the third and fifth decades of life. Almost 90% of leiomyomas are reported in this age group. Usually leiomyoma is encountered as a solitary mass which can multiply in 3–10% of cases.⁸ Eighty per cent of leiomyomas are intramural in the muscularis propria, with 10% arising in the proximal oesophagus, 30% in the middle oesophagus and 60% in the lower oesophagus.⁹ A majority of patients are asymptomatic. Dysphagia, chest pain and retrosternal discomfort appear when the tumour diameter is more than 4.5 cm.¹⁰ Radiological and endoscopic ultrasound examinations are useful diagnostic modalities. However, functional causes of dysphagia should be excluded by manometry and pH studies. Morris *et al*¹¹ reported

the first case of diffuse leiomyomatosis with dense eosinophilic infiltration. Eosinophils represent a minor component of inflammatory infiltrate with angiocentric distribution. Three cases of leiomyoma associated with eosinophilic esophagitis have been reported by Nicholson *et al*.¹² To the best of our knowledge, this is the second case of a solitary giant leiomyoma with eosinophilic infiltrates. The first case reported in 2011 by Linde and DiMaio *et al*¹³ was of a small 3×2.5 cm leiomyoma. Haematopoietic elements rarely infiltrate oesophageal leiomyoma but have been reported in idiopathic oesophagitis, diffused oesophageal leiomyomatosis and uterine tumours.¹⁴ The clinical significance of eosinophilic infiltration in oesophageal leiomyoma is not well understood. Probably, eosinophils play the same role in oesophageal smooth muscle regulation by releasing inflammatory mediators as in uterine smooth muscle

Figure 2 (A) Macroscopic picture after excision of an 8 cm oesophageal leiomyoma. (B) Abundant eosinophils are present within the muscle sheets.

regulation during pregnancy.¹⁵ Leiomyomas are treated according to the location and the size of the tumour. Small pedunculated leiomyomas can be treated with endoscopic band ligation or resected endoscopically.¹⁶ Another treatment option is video-assisted thoracoscopic or laparoscopic transhiatal enucleation.^{17–18} The transthoracic approach is suitable for large tumours over 5 cm, as in our case.

Learning points

- ▶ Leiomyomas are rare benign tumours of the oesophagus.
- ▶ CT scan of the thorax, barium swallow and endoscopy/ultrasound are useful diagnostic modalities.
- ▶ Complete excision through thoracotomy without injury to the oesophageal mucosa is the gold standard treatment for leiomyomas over 5 cm.
- ▶ Eosinophilic infiltration in oesophageal leiomyoma is a very rare finding. Eosinophils probably play the same role in the regulation of smooth muscles of the oesophagus as they do in the uterus.

Contributors HM wrote the case report. IC was the chief thoracic surgeon and he wrote the discussion. MA-A was the surgical resident who helped with care and images. MA was the consultant anaesthetist involved in this case and wrote part of the discussion.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Cox MA, Cooper BT, Sagar G. Endoscopy and computed tomography in the diagnosis and follow up of oesophageal leiomyoma. *Gut* 1995;37:288–91.
- 2 Lewin KJ, Appelman HD. Atlas of tumor pathology: tumors of the esophagus and stomach. Electronic fascicle V 2. Armed Forces Institute of Pathology, Washington, DC.
- 3 Lee LS, Singhal S, Brinster CJ, *et al.* Current management of esophageal leiomyoma. *J Am Coll Surg* 2004;198:136–46.
- 4 Healed J, Moussalli H, Hasleton PS. Diffuse leiomyomatosis of the esophagus. *Histopathology* 1986;10:755–9.
- 5 Appelman HD, Helwig EB. Gastric epithelioid leiomyoma and leiomyosarcoma. *Cancer* 1976;38:708–28.
- 6 Ranchod H, Kempson RL. Smooth muscle tumors of the gastrointestinal tract and retroperitoneum, a pathological analysis of 100 cases. *Cancer* 1977;39:255–62.
- 7 Kimura H, Konishi K, Kwawamura T, *et al.* Smooth muscle tumors of the esophagus: clinic pathological findings in six patients. *Dis Esophagus* 1999;12:77–81.
- 8 Mutrie CJ, Donahue DM, Wain JC, *et al.* Esophageal leiomyoma: a 40-year experience. *Ann Thorac Surg* 2005;79:1122–5.
- 9 Peters JH, DeMeester TR. Esophagus and diaphragmatic hernia. In: Brunicaudi FC, Anderson KD, Billiar RT, Dunn LD, Hunter GC, Pollock RE. eds *Schwartz's principles of surgery*. 8th edn. New York: McGraw-Hill, 2005:906.
- 10 Chak A. EUS in submucosal tumors. *Gastrointest Endosc* 2002;56:S43–8.
- 11 Morris CD, Wilkinson J, Fox D, *et al.* Diffuse esophageal leiomyomatosis with localized dense eosinophilic infiltration. *Dis Esophagus* 2002;15:85–7.
- 12 Nicholson AG, Li D, Pastorino U, *et al.* Full thickness eosinophilia in esophageal leiomyomatosis and idiopathic eosinophilic esophagitis. A common allergic inflammatory profile? *J Pathol* 1997;183:233–6.
- 13 Linde EM, DiMaio DJ. Solitary esophageal leiomyoma with eosinophilic infiltrate: case report and review of the literature. *Dis Esophagus* 2011;24:E5–7.
- 14 Vang R, Medeiros LJ, Samoszuk M, *et al.* Uterine leiomyomas with eosinophils: a clinicopathologic study of 3 cases. *Int J Gynecol Pathol* 2001;20:239–43.
- 15 Drury L, Sheppard BL, Bonnar J. The ultrastructure of mast cells in the uterus throughout the normal menstrual cycle and the postmenopause. *J Anat* 1991;175:51–63.
- 16 Sun S, Jin Y, Chang G, *et al.* Endoscopic band ligation without electrosurgery: a new technique for excision of small upper-GI leiomyoma. *Gastrointest Endosc* 2004;60:218–22.
- 17 Priego P, Lobo E, Alonso N, *et al.* Surgical treatment of esophageal leiomyoma: an analysis of our experience. *Rev Esp Enferm Dig* 2006;98:350–8.
- 18 Obuchi T, Sasaki A, Nitta H, *et al.* Minimally invasive surgical enucleation for esophageal leiomyoma: report of seven cases. *Dis Esophagus* 2010;23:E1–4.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow