

CIRUGÍA ESPAÑOLA

www.elsevier.es/cirugia



Original article

Leiomyoma of the oesophagus[☆]

Luis F. Loviscek,^{a,*} Jong Hyoun Yun,^a Yoo Sun Park,^a Ariel Chiari,^a Cristian Grillo,^a and María C. Cenoz^b

^aSección de Cirugía Esofágica, Departamento de Cirugía, Hospital Pirovano, Ciudad de Buenos Aires, Argentina

^bAnatomía Patológica, Hospital Pirovano, Ciudad de Buenos Aires, Argentina

ARTICLE INFORMATION

Article history:

Received May 21, 2008

Accepted September 1, 2008

On-line February 5, 2009

Keywords:

Oesophagus

Benign tumour

Leiomyoma

Surgery

Thoracoscopy

A B S T R A C T

Introduction: Oesophageal leiomyoma is a rare tumour. The purpose of this paper is to describe the form of presentation, the approaches and the surgical technique, in 5 consecutive cases.

Material and method: A retrospective analysis was performed on the data recorded prospectively on 5 consecutive patients operated on between 2003 and 2007.

Results: All the patients were seen due to dysphagia to solids. All were examined with contrast radiology, video-endoscopy, and computed tomography, although 4 of them also had ultrasound endoscopy performed. The surgical technique was extra-mucosal enucleation, with 3 cases by thoracoscopy and 2 by thoracotomy. There were no complications. The immunohistochemistry study was negative for CD117 and CD34 in all cases.

Conclusions: Enucleation of oesophageal leiomyomas by video-thoracoscopy is safe, well tolerated, and there is a rapid recovery, and is the procedure of choice for this benign tumour. Thoracotomy may be necessary in some presentations and locations.

© 2008 AEC. Published by Elsevier España, S.L. All rights reserved.

Leiomioma de esófago

R E S U M E N

Introducción: El leiomioma de esófago es un tumor poco frecuente. El objetivo de esta comunicación es describir la forma de presentación, las vías de abordaje y la técnica quirúrgica en 5 casos consecutivos.

Material y método: Se analizaron retrospectivamente los datos registrados prospectivamente de 5 pacientes consecutivos operados entre 2003 y 2007.

Resultados: Todos los pacientes consultaron por disfagia a sólidos. Todos fueron estudiados con radiología contrastada, videoendoscopia y tomografía computarizada, si bien en 4 de ellos también se realizó ecoendoscopia. La técnica quirúrgica fue la enucleación extramucosa: 3 casos por toracoscopia y 2 por toracotomía. No se observaron complicaciones. El estudio inmunohistoquímico fue negativo para CD117 y CD34 en todos los casos.

Palabras clave:

Esófago

Tumor benigno

Leiomioma

Cirugía

Toracoscopia

[☆]Study presented and read in the 78th Argentinean Conference of Surgery. Buenos Aires, October 2007.

*Author for correspondence.

E-mail address: lovisceklf@yahoo.com.ar (L.F. Loviscek).

0009-739X/\$ - see front matter © 2008 AEC. Published by Elsevier España, S.L. All rights reserved.

Conclusiones: La enucleación de los leiomiomas de esófago por videotoracoscopia es segura y bien tolerada, permite una rápida recuperación y es el procedimiento de elección para este tumor benigno. La toracotomía puede ser necesaria en algunas presentaciones y localizaciones.

© 2008 AEC. Publicado por Elsevier España, S.L. Todos los derechos reservados.

Introduction

Leiomyoma is a benign tumour. It is rare, 0.4%–1% of oesophageal tumours,¹ but it is the most common of benign tumours (67%–80%) of the oesophagus.² Its differential diagnosis should be established with cancer, gastrointestinal stromal tumours (GIST), and other benign tumours. It generally presents with an oval-shaped solitary intramural mass which originates in the muscularis propria and in an elongated, ring-shaped, horseshoe, or spiral form, surrounding the oesophagus wall.^{1,2} Leiomyomas can be multiple in approximately 5% of cases.³ Endoscopic appearance is that of a mass covered by normal mucosa.⁴ The need to resect or not a leiomyoma is based on the symptoms it produces, especially dysphagia, and on the differential diagnosis with GIST.⁵ The most used technique is extramucosal enucleation⁶.

The objective of this study is to describe symptoms, form of presentation, surgical prescription, approach, and surgical technique used for 5 patients with oesophageal leiomyoma treated surgically.

Material and method

The medical records of 5 patients with oesophageal leiomyoma, operated between 2003 and 2007, were retrospectively analyzed. Data taken into account were, gender, main symptoms, diagnostic studies done, location, approach, surgical technique, evolution, and anatomic-pathological study of the resected tumour. Of the 5 patients, 3 were males. Average age was 51 (48–57) years. In all cases, the technique used was extramucosal enucleation, by right thoracotomy in 2 cases and by videothoracoscopy (VT) in 3.

With patients operated by VT, the technique employed was the following: patient in left lateral position with general anaesthesia and selective bronchial intubation. The surgeon was located to the right of the patient. Previous pulmonary collapse, 4 trocars were placed in the right haemithorax, on mid-axillary, anterior axillary and posterior axillary lines. In all cases, 30° optics was used, inserted by a 10 mm trocar placed on the mid-line of the seventh and eighth intercostal space for high tumours, and in the fourth and fifth intercostal space for low tumours. A 5 mm trocar for inserting a grasper and another 5 mm trocar for inserting the hook, scissors or 5 mm Ligasure® forceps, on anterior and posterior axillary lines. A fourth 10 mm trocar was used to insert the pulmonary retractor. In one case a fifth 10 mm trocar was used. Once vision and the surgical area were established, the collapsed lung was separated to show the oesophagus, incising if necessary the triangular ligament. Next, the mediastinal

pleural cavity was incised, and the oesophagus was released at the height of the tumour. The azygos vein was conserved in all cases. Oesophageal muscle fibres were incised and separated, with particular care for preserving vagus nerves and/or their branches, until the tumour was exposed, and then extramucosal enucleation of the leiomyoma was carried out using blunt dissection techniques. In some cases, placing a stitch in the leiomyoma allowed retracting of the tumour and facilitated enucleation. Once the leiomyoma was enucleated and the security of the oesophageal mucosa was confirmed, the oesophageal muscle fibres were reconnected with silk stitches 2/0. In all cases, 1 or 2 thoracic tubes were inserted.



Figure 1 - Regular image with clear edges which form an acute angle with the oesophageal wall.

Results

All patients were consulted for dysphagia of solids. Two out of 5 patients suffered from non-cardiogenic thoracic pain (NCTP). All were studied using contrast radiology, videoendoscopy (VE), and computed tomography (CT). In 4 cases, the study was completed with an echoendoscopy. The topographical distribution of the lesions was the following: superior thoracic, 1 case; mid-oesophagus, 3; and inferior intrathoracic oesophagus, 1. In 3 of 5 cases, the lesion was nodular (Figure 1), while the 2 remaining cases were spiral ring-shaped (Figure 2). In all cases, the endoscopic image was of a submucosal lesion covered by normal mucosa. Biopsy was not taken in any cases. In all cases, the CT showed a thickening of the oesophageal muscle (Figure 3). The echoendoscopy revealed regular tumoural mass in relation to the muscularis propria (Figure 4). The 3 leiomyomas with nodular appearance were resected by VT. Two cases with spiral ring-shaped manifestation, one of them with superior thoracic localization in relation to the large vessels, were resected by thoracotomy. The intraoperative VE was only used



Figure 2 - Irregular image which corresponds to a spiral-shaped leiomyoma.



Figure 3 - Thickening of the oesophageal wall.

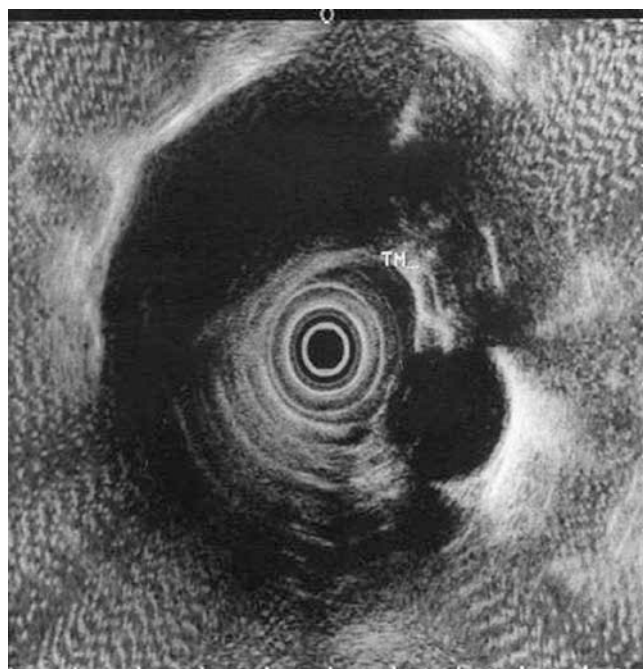


Figure 4 - Hypoechoic and homogeneous regular mass dependent on muscularis propria.

Pathological anatomy

Histology of incised sections showed a tumour of mesenchymal origin, composed of spindle cells with eosinophilic cytoplasm and nuclei that were oval, elongated, with characteristic blunt edges, laid out in interwoven fascicles accompanied by mild infiltrate of lymphocytes, plasmocytes, and eosinophils. Atypical mitosis or necrosis areas were not found in any case. In the 5 cases, immunoperoxidase technique was carried out for demonstrating vimentin, desmin, HHF-35, CD34, and CD117, by using monoclonal and polyclonal serum as an avidin-biotin detection system and as a diaminobenzidine reveler. Vimentin, desmin, and HHF-35 were positive. In all cases, immunohistochemistry was negative for CD117 and CD34.

Table – Symptoms, location, macroscopy, and immunohistochemistry

Sex/age	Symptoms	VE	Treatment	Macroscopy	c-kit CD117 and CD34 ^a
Female/50	Dysphagia	To 28 cm	Right thoracotomy	Ring-shaped in horseshoe 3×2 cm	Negative
Male/49	Dysphagia	To 30 cm	Right thoracoscopy	Nodular 4×3 cm	Negative
Male/48	Dysphagia	To 31 cm	Right thoracoscopy	Nodular 5×4 cm	Negative
Male/57	Dysphagia, NCTP	To 35 cm	Right thoracoscopy	Nodular 4×3 cm	Negative
Female/51	Dysphagia, NCTP	To 26 cm	Right thoracotomy	Spiral 5×2.5 cm	Negative

NCTP indicates non-cardiogenic thoracic pain; VE, videoendoscopy.
^aMarkers for gastrointestinal stromal tumours.

Discussion

Leiomyoma, although rare, is the most common benign tumour of the oesophagus. Dysphagia is the most frequent symptom, followed by thoracic pain.⁵ The majority develop in the bottom third of the oesophagus.⁷ Four of our 5 cases were localized below the carina (Table). Currently, leiomyomas are recognized as different benign tumours from GIST. Leiomyoma has a radiological appearance characteristic of concave images and clear edges.¹ By endoscopy, these tumours appear as mobile submucosal masses covered by normal mucosa⁴; endoscopic biopsy is not recommended because adhesion can occur from mucosa to the tumour and complicate extramucosal enucleation during surgical procedure.^{8,9} Perforation of the tumoural mass with a fine needle by endosonography or CT, although debated, is used in some centres for carrying out preoperative diagnosis and ruling out GIST.¹⁰ In cases of ulcerated mucosa and suspicion of cancer, endoscopic biopsy is prescribed.⁹ Endosonography is a great help for determining the location and depth of the tumour, and it is considered the method with the greatest diagnostic specificity for leiomyoma.⁷ CT is useful for determining the tumour's size and its relationship with neighbouring organs. Small leiomyomas, generally incidentalomas, can be controlled periodically by radiography and endoscopy with no resection, given its slow growth.^{2,4} Symptoms and suspicion of GIST strengthen surgical prescription. The first extramucosal enucleation of an oesophageal leiomyoma was described by Ohsawa in 1933. Since then, thoracotomy procedure was standard. In 1992, enucleations by videothoracoscopy were published for the first time^{11,12}; currently, this procedure is considered the surgical treatment of choice for oesophageal leiomyomas. In leiomyomas of the inferior third section, especially those of the intra-abdominal oesophagus or in those distal of 4–5 cm, enucleation by laparoscopy is a valid option.¹³ VT is the preferred technique due to its referred advantages with respect to post-thoracotomy pain, fast recovery, and pulmonary complications (atelectasis, pleural effusion, etc).³ Thoracotomy for large leiomyomas or for spiral-shaped tumours gives greater security in complete resection. Resection is safer, especially in leiomyomas connected to large vessels, and reconstruction of muscular layers of the oesophagus is simpler in these cases.^{14,15} With respect

to this point, it is necessary to assert that reconstruction of the oesophageal muscle is important for preventing mucosal prolapse,^{2,8} although some authors have observed that long extramucosal defects can be left without necessarily developing a pseudodiverticulum. In all of our cases, we have reconstructed the muscular layer of the oesophagus, covering the surgical opening. The intraoperative VE can be useful for localizing the lesion in small leiomyomas, for identifying the border between oesophageal mucosa and leiomyoma during dissection, and/or for confirming the mucosa's security while completing enucleation.^{3,5} In this series, we used enucleation in one single case. The most common causes of conversion during an enucleation attempt by VT are technical difficulties which can be manifested with spiral or helical shaped leiomyoma, mucosal lesion, and the patient's intolerance due to pulmonary collapse.

Endoscopic resection of leiomyoma has been described,¹⁶ but it should be limited to particular cases in which only the muscularis propria is affected on its internal layer. Oesophageal resection may be prescribed for giant leiomyomas with a highly affected esophageal wall.⁹ The anatomopathological study with immunohistochemistry is obligatory for ruling out GIST. Different from GIST, leiomyomas are not immunoreactive for c-kit CD117 and CD34.^{17,18}

REFERENCES

- Shamji F, Todd TR. Benign tumors. In: Pearson FG, Hiebert CA, Deslauries J, et al, editors. *Oesophageal surgery*. New York: Churchill Livingstone; 1995. p. 519–53.
- Mutrie Ch, Donahue DM, Wain JC, Wright CD, Gaisert HA, Grillo HC, et al. Oesophageal leiomyoma: A 40-year experience. *Ann Thorac Surg*. 2005;79:1122–5.
- Peracchia A, Bonavina L, Bardini R, Montorsi M, Segalin A. Thoracoscopic enucleation of esophageal leiomyoma. In: Peters J, DeMeester T, editors. *Minimally invasive surgery of the foregut*. St Louis: QMP; 1994. p. 239–44.
- Rice TW. Benign esophageal tumors: esophagoscopy and endoscopic esophageal ultrasound. *Semin Thorac Cardiovasc Surg*. 2003;15:20–6.
- Zaninotto G, Portale G, Constantini M, Rizzetto C, Salvador R, Rampado S, et al. Minimally invasive enucleation of esophageal leiomyoma. *Surg Endosc*. 2006;20:1904–8.
- Ohsawa T. Surgery of the esophagus. *Arch Jpn Chir*. 1933;10:605–8.

7. Xu GQ, Zhang BL, Li YM, Chen LH, Ji F, Chen WX, et al. Diagnostic value of endoscopic ultrasonography for gastrointestinal leiomyoma. *World J Gastroenterol*. 2003;9:2088-91.
8. Bonavina L, Segalin A, Rosati R, Pavanello M, Peracchia A. Surgical therapy of esophageal leiomyoma. *J Am Coll Surg*. 1995;181:257-62.
9. Punpale A, Rangole A, Bhambhani N, Karimundackal G, Desai N, Ashwin de Souza, et al. Leiomyoma of esophagus. *Ann Thorac Cardiovasc Surg*. 2007;13:78-81.
10. Ando N, Goto H, Niwa Y, et al. The diagnosis of GI stromal tumours with EUS-guided fine needle aspiration with immunohistochemical analysis. *Gastrointest Endosc*. 2002;55:37-43.
11. Bardini R, Segalin A, Ruol A, Pavanello M, Peracchia A. Videothoracoscopic enucleation of esophageal leiomyoma. *Ann Thorac Surg*. 1992;54:576-7.
12. Everitt NJ, Glinatsis M, McMahon MJ. Thoracoscopic enucleation of leiomyoma of the oesophagus. *Br J Surg*. 1992;79:643.
13. Marmuse JP, Johanet H, Louvent F, Benhamou G. Leiomyomas of the lower third of the esophagus. Value of transhiatal enucleation. *Ann Chir*. 1994;48:446-51.
14. Kent M, D'Amato T, Nordman C, Schuchert M, Landreneau R, Alvelo-Rivera M, et al. Minimally invasive resection of benign esophageal tumors. *J Thorac Cardiovasc Surg*. 2007;134:176-81.
15. von Rahden BH, Stein HJ, Feussner H, Siewert JR. Enucleation of submucosal tumors of the esophagus: minimally invasive versus open approach. *Surg Endosc*. 2004;18:924-30.
16. Shin CS, Jung IS. Endoscopic removal of submucosal tumors: preprocedure diagnosis, technical options, and results. *Endoscopy*. 2005;37:646-54.
17. Logroño R, Jones DV, Faruqi S, Bhutani MS. Recent advances in cell biology, diagnosis, and therapy of gastrointestinal stromal tumor (GIST). *Cancer Biol Ther*. 2004;3:251-8.
18. Miettinen M, Virolainen M, Maarit-Sarlomo-Rikala. Gastrointestinal stromal tumors value of CD34 antigen in their identification and separation from true leiomyomas and schwannomas. *Am J Surg Pathol*. 1995;19:207-16.