Leiomyoma of the oesophagus managed by thoracoscopic enucleation

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ABSTRACT

The authors document two patients with oesophageal leiomyoma. In the first patient, a 41-year-old man, enucleation of the oesophageal leiomyoma was initially attempted by a thoracoscopic approach, but because of adherence of the tumour to the oesophageal mucosa, enucleation was completed by thoracotomy. Thoracoscopic enucleation was successfully performed in the second patient, a 62-year-old man. This paper includes a literature review on the pathology, diagnosis and surgical approach in the management of oesophageal leiomyoma. In conclusion, prudent use of thorascopic approach in the enucleation of oesophageal leiomyoma could potentially result in shorter hospital stay, decreased postoperative pain and reduced requirement for postoperative analgesia.

Keywords: **endoscopic ultrasonographic fine-needle aspiration biopsy, gastrointestinal stromal tumour, leiomyoma, oesophageal leiomyoma, thoracoscopy, thoracoscopic enucleation.**

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INTRODUCTION

Oesophageal leiomyomas are uncommon tumours[1-3]. Malignant transformation is extremely rare, but removal is often required in symptomatic patients with dysphagia, retrosternal discomfort or bleeding[2,4,5]. In leiomyomas of the thoracic oesophagus, open transthoracic extramucosal enucleation has traditionally been the standard surgical treatment[2,6]. Since 1992, thoracoscopic enucleation, with its advantages of reduced hospital stay, pulmonary complications and thoracotomy pain, has been reported[4,6-8]. However, there has not been any report of thoracoscopic enucleation of oesophageal leiomyomas being performed in Singapore. This report presents the experience of the thoracoscopic approach in the enucleation of oesophageal leiomyoma performed on two patients by one of the authors (TKT) at the National University Hospital, Singapore, and includes a review of the literature.

CASE REPORTS

Case 1

In 2001, a 41-year-old Bangladeshi man presented with retrosternal discomfort especially after meals. He was otherwise asymptomatic. Chest radiograph, electrocardiography and biochemical tests (including haemoglobin, urea, creatinine and electrolyte levels), were normal. Oesophagogastroduodenoscopy (OGD) showed a rounded submucosal mass at the mid-oesophagus. Endoscopic ultrasonography confirmed a well-defined 2.5 cm × 3.3 cm mass. There was no lymphadenopathy in the paraoesophageal region and the coeliac axis. Endoscopic ultrasonographic fine-needle aspiration biopsy (EUS-FNAB) showed a few cell clusters with occasional spindle shaped nuclei suggestive of benign non-epithelial tumour.

Right thoracoscopy showed a firm mural mass in the oesophageal wall, located 30-35 cm from the incisors. Initial dissection to expose the tumour by diathermising the pleura and blunt dissection followed by splitting muscularis propria was performed. As the tumour appeared to be large and adherent to the mucosa, surgery was converted to open thoracotomy. An elongated, smooth-surfaced 5.0 cm × 2.5 cm × 1.5 cm well-encapsulated, greyish-white tumour was removed after separating the muscularis propria fibres, taking special care to avoid mucosal laceration. Oesophageal muscle and pleura were then apposed with interrupted sutures. The elongated, homogeneous tumour mass was sectioned to show a well-demarcated leiomyoma, with focal, spotty calcification. There was no evidence of malignancy detected histologically. Immunohistochemistry was not routinely performed at that time. In the follow-up period, the patient was

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well except for some right chest wall postoperative pain and numbness for six weeks.

Case 2

In 2003, a 62-year-old Chinese man presented with recurrent dysphagia for four years and retrosternal discomfort of two years duration. He experienced difficulty swallowing solid food. There was accompanying non-bilious vomiting. The patient was previously diagnosed to have hiatus hernia and reflux oesophagitis. OGD showed erosive oesophagitis as well as a low-grade stricture, possibly due to an extrinsic lesion compressing the mid-oesophagus. Barium swallow confirmed extrinsic indentation of the oesophagus at the level of the carina. Computed tomography showed a bulky intrinsic oesophageal lesion arising from its wall, causing a sudden change in its lumen size, at the level of the carina. No lymphadenopathy was detected. Although the EUS-FNAB material was insufficient to make a definitive diagnosis, it showed that the lesion was composed of stromal fragments, described as bands of fibres with bland spindly nuclei. No lymphoid cells were seen.

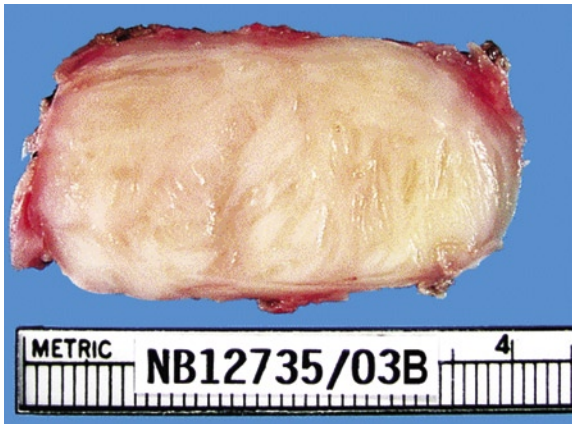

Fig. 1 Specimen photograph of the resected leiomyoma shows a relatively-circumscribed firm whitish nodule. The cut surface shows a whitish whorled appearance without gross necrosis, haemorrhage or cyst formation.

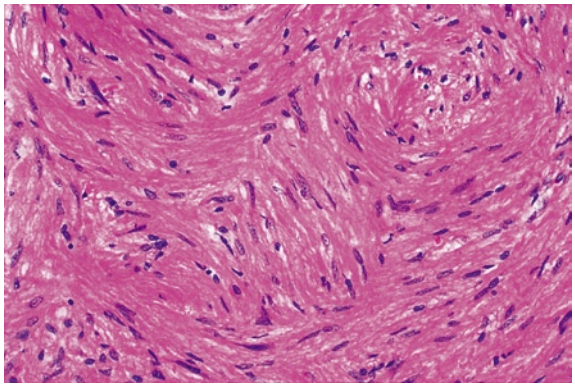

Fig. 2 Photomicrograph shows no significant pleomorphism, mitotic activity, necrosis, haemorrhage or cystic degeneration (Haematoxylin & eosin, ×40).

During surgery, the patient was placed in the right lateral position, deflating his right lung using a double-lumen endotracheal tube. Five thoracoscopic ports were inserted and an OGD was performed to assist in localising the tumour. The azygos vein was freed and transected with a gastrointestinal anastomosis (GIA) vascular stapler. This facilitated dissection of the mid-oesophagus bearing the tumour. After the muscle over the tumour was split, the well-encapsulated tumour was separated from the surrounding oesophageal mucosa by blunt dissection and diathermy. Special precautions were made when freeing the tumour from the oesophageal mucosa so as to avoid damaging it. The split muscularis propria was then closed with interrupted sutures. OGD confirmed that there was no breech of the oesophageal mucosa.

The benign-looking tumour measured 4.3 cm × 2.2 cm × 2.1 cm (Fig. 1). The firm, whitish, whorled nodule did not have any gross necrosis, haemorrhage or cyst formation. Frozen section was suggestive of a leiomyoma. Paraffin section confirmed a moderately cellular spindle cell neoplasm with no significant pleomorphism, mitotic activity, necrosis, haemorrhage or cystic degeneration (Fig. 2). Immunohistochemistry demonstrated the expression of the desmin antibody but was negative for c-kit (CD117) and CD34, which favoured the diagnosis of a leiomyoma rather than a gastrointestinal stromal tumour (GIST). A water-soluble contrast swallow done on the fifth postoperative day showed free flow of contrast from oesophagus to stomach, with no leakage. Recovery was uneventful, although the patient had right-sided chest wall pain for three months.

DISCUSSION

Although patients with oesophageal leiomyoma may be asymptomatic, many patients, including our two cases, complain of dysphagia and vague retrosternal pain[5]. Other symptoms include breathlessness, epigastric discomfort and regurgitation. Bleeding may also occur if the overlying mucosa is ulcerated from coexisting reflux oesophagitis[2]. Leiomyomas are now known to be benign tumours distinct from GISTs[9-11]. Unlike leiomyomas, GISTs are immunoreactive for c-kit protein (CD117) and CD34. Expression of both these proteins, tumour size exceeding 5 cm and a mitotic rate of over 5/50 high power field (HPF) are predictive of malignant behaviour[12].

In the management of a submucosal tumour, preoperative distinction between a leiomyoma and a GIST can be made with EUS-FNAB[13,14]. However,

it is debatable whether EUS-FNAB should be routinely used. Some studies have reported that in patients who have undergone FNAB, there was an increase in the incidence of mucosal tears during enucleation[7,15,16]. We believe that preoperative EUS-FNAB should be recommended, particularly in the management of larger submucosal tumours. Malignant behaviour is common in GIST larger than 5 cm, and resection rather than enucleation would be the appropriate surgical treatment in these patients. Symptomatic oesophageal leiomyomas are successfully treated by enucleation. It has been suggested that tumours up to 8 cm can be safely enucleated without significant postoperative dysphagia, as long as the mucosa is intact and the myotomy is reapproximated[6,7,17,18]. Although larger lesions can be removed via enucleation, the resulting defects are too large to allow a tension-free suture, requiring a tissue flap to prevent mucosal bulging[7,19].

Enucleation of oesophageal leiomyoma has traditionally been performed by open thoractomy[2]. Since 1992, the thoracoscopic approach has been increasingly performed[4,20]. The thoracoscopic approach offers advantages of being less invasive, and avoiding the scarring and discomfort of thoracotomy[1,4]. Operating time was reported to be similar (mean 95 min) for both thoracoscopic and open approaches[8]. The thoracoscopic approach allows rapid, full re-expansion of the lung with minimal adverse effects on pulmonary function, such as a lesser degree of pulmonary atelectasis, dystelectasis and pleural effusions[4,6,7]. Shorter hospital stay (6.8 days versus 10.2 days), decreased postoperative pain and reduced requirement for postoperative analgesia were also noted[7,8].

Nevertheless, the reduction of postoperative pain after thoracoscopy has not been substantiated by other workers[21], and our second patient also experienced considerable discomfort for three months. It has been suggested that excessive torquing of the trocars in the intercostal space, direct injury to the neurovascular bundle by electrosurgical access, or blunt dissection can result in chronic pain[22,23]. Richardson and Sabanathan recommended the resection of a rib eclipse with a "Sari" punch, and the alignment of the instruments along one intercostal space to reduce the likelihood of peripheral nerve damage, thereby reducing chest wall pain[22]. In summary, minimally- invasive surgery by the thoracoscopic approach appears to have some advantages over the open thoracotomy approach. A thoracoscopic approach increases the repertoire of surgical techniques and when appropriately applied, can be beneficial in the enucleation of oesophageal leiomyoma.

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