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**Case Report**

# Thoracoscopic management of a giant horseshoe shaped oesophageal leiomyoma: a case report

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## ABSTRACT

Benign oesophageal tumours are considered rare, and most common are leiomyomas with the incidence of 0.005%. Many cases are asymptomatic, up to 15%–50%, and most cases are discovered incidentally. These may vary in size but are typically smaller than 3 cm. Giant esophageal leiomyoma is defined as tumour greater than 10 cm in diameter. Traditionally, open thoracotomy and enucleation have been the primary treatment modalities for oesophageal leiomyoma. However, in recent years, video-assisted thoracoscopic surgery (VATS) enucleation has gained recognition for its proven advantages as a minimally invasive surgical option. Herein we present our experience with patient presenting with cough rather than dysphagia as a main symptom, who was diagnosed to be having giant oesophageal leiomyoma. VATS guided enucleation was accomplished successfully. Size of lesion was 10.0×8.0×7.0 cm. Postoperative recovery was uneventful.

**Keywords:** Enucleation, Video-assisted thoracoscopic surgery, Giant leiomyoma, Oesophageal tumour

## INTRODUCTION

Benign oesophageal lesions as such are rare, among which the most common are leiomyomas with an incidence of 0.005%.[1] Oesophageal leiomyomas arise from smooth muscle, may differ in size but are usually less than 3 cm in size. Giant oesophageal leiomyomas are defined as lesions that are greater than 10 cm, the incidence of these has been reported as 17%.[2] This affects patients between 20 and 50 years of age, with male to female ratio of 2: 1.[3] This tumour is discovered as an incidental finding on imaging in 15–50% of the cases, as they are predominantly asymptomatic during presentation due to the small size.

However, larger lesions may impede on local structures causing obstructive symptoms such as dysphagia.[4] Surgical treatment is typically only recommended for leiomyomas that are symptomatic or larger than 5 cm. The traditional surgical method for treating oesophageal leiomyoma is an open thoracotomy followed by extra-mucosal blunt enucleation. Over time, this approach has been largely replaced by minimally invasive thoracoscopic techniques, such as video-assisted thoracoscopic surgery (VATS).[5] Here we present a case of a 36-year-old male patient with a giant oesophageal leiomyoma that was treated with VATS.

## CASE REPORT

A 36-year-old male patient presented with cough for one month to a general physician, where he was evaluated. He had no complaints of dysphagia, nausea, loss of weight, or upper abdominal pain. Patient did not have relevant personal and family history and was a non-smoker.

Physical examination was unremarkable, and all baseline blood investigations were within normal limits. CT thorax was done, which showed a well-defined soft tissue

mass measuring 5.4×7.5×7.8 cm in the mediastinum, arising from the oesophagus above the gastro-oesophageal junction, with significant compression of lower oesophagus GIST. An endoscopic ultrasound (EUS) showed a well demarcated heteroechoic lesion of size 7.4×5.8 cm arising from 4th layer of oesophagus.

Fine needle aspiration was then performed which was reported as Benign spindle cell neoplasm. Immunohistochemistry was positive for SMA, negative for C-KIT, S100, and CD34. Pathology confirmed the diagnosis of leiomyoma of the oesophagus, distinguishing the leiomyoma from a gastrointestinal stromal tumour (GIST).

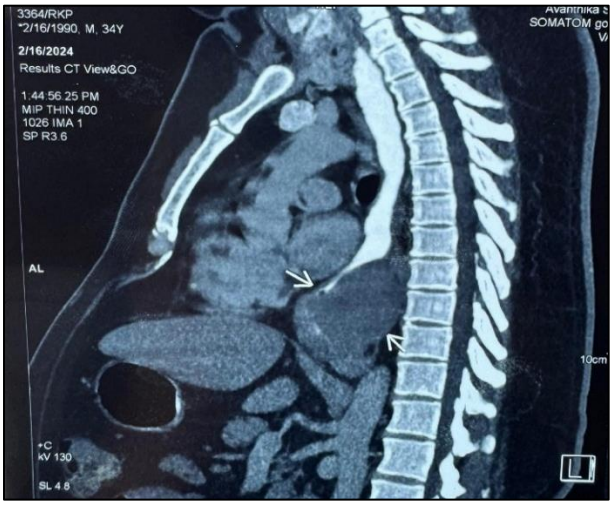


**Figure 1: CT thorax sagittal section showing well defined soft tissue mass abutting descending aorta.**

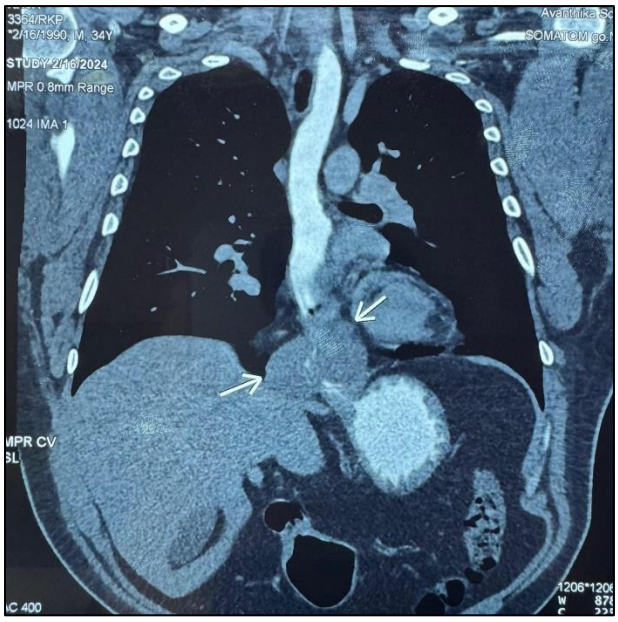


**Figure 2: CT thorax coronal section showing the mass lesion just above the gastro-oesophageal junction causing significant compression of lower oesophagus with free passage of oral contrast into stomach.**

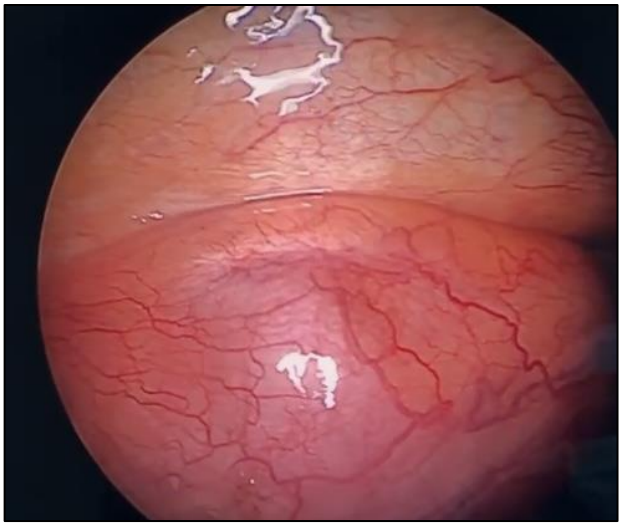

**Figure 3: Intraoperative image from thoracoscopy showing the oesophageal mass.**

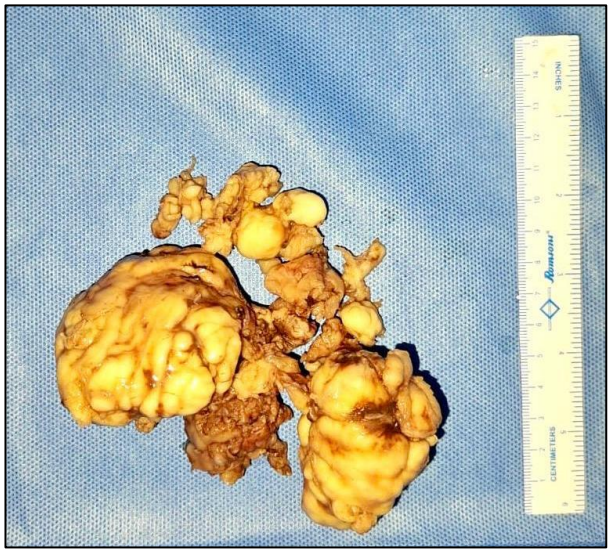

**Figure 4: Excised specimen of oesophageal leiomyoma immediately after operation.**

7 months after initial presentation, patient was taken up for Right VATS and Enucleation of oesophageal Leiomyoma under general anaesthesia. Intraoperatively the lesion was found to be much larger and was almost circumferential at the lower oesophagus. After meticulous dissection, the mass was excised in toto. Intraoperative assistance was sought from cardiothoracic surgeons and specimen was extracted via posterolateral mini-thoracotomy.

The excised mass was horseshoe shaped, 10 cm at the largest dimension (10.0×8.0×7.0 cm). An intraoperative upper GI endoscopy was performed to ensure adequate lumen patency and to inspect the mucosal integrity through direct visualization. Insufflation tests showed no mucosal leaks, and there were no surgical complications. ICD was placed, which was removed on POD10. His

post-operative course was unremarkable, he was started on liquid diet on POD1 and slowly diet was stepped up. He is currently doing well without symptoms and is tolerating a regular diet. Histopathological report mentioned presence of interlacing fibers of smooth muscle cells arranged in whorled appearance without any evidence of malignancy suggestive of leiomyoma of oesophagus.

## DISCUSSION

Morgagni first described leiomyoma in 1761, while Munro was the first to report a localized leiomyoma in the oesophageal wall in 1797.[6] Oesophageal leiomyoma is the most common benign tumour originating from the smooth muscle cells of the oesophagus. It can occur in any segment of the oesophagus, but it is reported to affect the distal third in 60% of cases, the middle third in 30%, and the upper third in 10% of cases.[7] This distribution reflects the relative abundance of smooth muscle cells present along the oesophagus. It is a slowly growing intramural tumour with very limited malignant potential. The reported size of oesophageal leiomyomas ranges from 1 to 30 cm.[8] Leiomyomas larger than 10 cm are classified as giant oesophageal leiomyomas.[9]

Oesophageal leiomyoma often presents with no specific symptoms, and diagnosis is frequently an incidental finding. While the clinical presentation can vary based on the tumour's size and location, no consistent association has been established between these factors. Shin et al, reported one of the largest series of oesophageal leiomyomas, with clinical presentations ranked as follows: asymptomatic (58%), dysphagia (12%), epigastric discomfort (8%), dyspepsia (6%), chest discomfort (2%), and regurgitation (1%).[10] Rare features may include bleeding and weight loss. Dysphagia typically occurs when the tumour's diameter exceeds 5 cm. Notably, our patient presented primarily with cough, which has rarely been reported as a predominant or sole symptom of oesophageal leiomyoma. Larger oesophageal leiomyomas often grow outward from the oesophageal lumen, meaning that dysphagia may not necessarily correlate with the tumour's size in such cases.

Preoperative diagnosis of oesophageal leiomyoma is often a challenge. As in our case, it can present as an incidental radiologic finding. Esophagoscopy will show normal mucosa and submucosal lesion. Computed tomography (CT) and endoscopic ultrasound (EUS) are very valuable in making diagnosis, they will delineate the intramural nature of tumour without any mediastinal lymphadenopathy. In CT images, a leiomyoma presents as a homogenous mass within the oesophageal wall, showing no signs of invasion. While it can be challenging to distinguish leiomyomas from other oesophageal tumours, the presence of calcifications is nearly pathognomonic for leiomyoma.[11] Endoscopic ultrasound is highly accurate in diagnosing oesophageal leiomyomas, revealing a hypoechoic, homogeneous mass with sharply defined margins in the muscular layer.[12] Preoperative FNA diagnosis helps confirm the benign nature of the lesion however care must be ensured to avoid mucosal damage. This patient's leiomyoma was larger than most, as oesophageal leiomyomas exceeding 5 cm are relatively uncommon. This case report contributes to the growing body of evidence that VATS can be employed for the enucleation of larger leiomyomas. It also highlights the usefulness of simultaneous endoscopic visualization in challenging locations, such as in this case where it was close to the gastroesophageal junction (GEJ). The use of VATS for enucleation of oesophageal leiomyoma was first described by Everitt in 1992.[13] However, VATS was not the preferred approach for the management of oesophageal leiomyoma over open thoracotomy until 2011.[5]

Over the years, the popularity of minimally invasive surgical management of leiomyomas has continued to increase. It has even been shown to be effective in treating giant oesophageal leiomyomas as in our case. However, the use of direct endoscopic evaluation via esophagoscopy during this patient's procedure provided additional visual and anatomical reassurance that the underlying oesophageal mucosa was neither disrupted nor narrowed during the mass enucleation. This case suggests that giant oesophageal submucosal masses (~10 cm) close to the GEJ can potentially be enucleated using a combined VATS and endoscopic approach.

When comparing VATS with open surgery, few previous studies have shown that patients undergoing VATS experience significantly shorter operative times, less blood loss, quicker time to oral intake, and shorter postoperative stays.[14] Additional studies have shown that VATS is associated with lower rates of intraoperative and postoperative complications compared to open thoracotomy.[5] In line with these findings, the patient presented in this report had an unremarkable surgical and post-operative course, was able to tolerate clear liquid diet on POD1.

## CONCLUSION

Here, we present a patient with a giant oesophageal leiomyoma that was enucleated using a combined endoscopic and right VATS approach. Occasionally, giant oesophageal leiomyoma of oesophagus may mimic respiratory tract pathology, with cough as a main symptom, so one needs to consider this possibility even in treating such patients empirically. The purpose of this report is to contribute to the limited literature on the minimally invasive surgical treatment of relatively large oesophageal leiomyomas. This case underscores that VATS combined with endoscopic visualization is an effective and safe option for treating giant oesophageal leiomyomas and can be associated with minimal risk.

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