



Uniportal Video-Assisted Thoracic Surgical Enucleation of a Giant Upper Esophageal Myxoid Leiomyoma: A Case Report

Yao X*, Li G, Song Y and Jiang L

Department of Thoracic Surgery, The Public Health Clinical Center of Chengdu, China

Abstract

Benign esophageal tumor is an uncommon disease of the esophagus. These tumors, often present as mediastinal lesions on Computed Tomography (CT) scans. Leiomyoma of the esophagus is the most common type of benign disease of the esophagus, and it usually occurs in the distal esophagus and/or gastroesophageal junction. In contrast, leiomyoma in the upper esophagus is rare and challenging to treat. Here, we report on a 61-year-old female patient with giant (defined as >10 cm diameter) myxoid leiomyoma in the proximal esophagus. The patient experienced dysphagia and dyspnea for about 3 months. Uniportal Video-Assisted Thoracic Surgery (VATS) involving enucleation of the leiomyoma was performed, followed by primary suture of the esophagus. The patient recovered smoothly and was discharged 7 days after the operation. A diagnosis of myxoid leiomyoma of the esophagus was made based on the postoperative pathological results. Uniportal VATS could be a safe and effective treatment approach for this benign disease. Consideration should be given to the diagnosis of giant myxoid leiomyoma when patients present with dysphagia and dyspnea with a giant mediastinal mass in the upper mediastinum.

Keywords: Myxoid esophageal leiomyoma; Uniportal video-assisted thoracic surgery; Enucleation; Benign esophageal tumor; Esophageal rupture

Introduction

Leiomyoma, a type of mesenchymal tumor, is rare, but it is the most common type of benign tumor of the esophagus [1,2]. Esophageal leiomyoma with a diameter >10 cm is defined as “giant esophageal leiomyoma”. Esophageal leiomyoma generally occurs in the distal esophagus and/or gastroesophageal junction, while leiomyoma in the upper esophagus is rare. Giant upper esophageal leiomyoma is difficult to distinguish from primary posterior tumors. Misdiagnosis of this benign tumor may lead to unnecessary esophagectomy, so correct diagnosis is important to avoid this. Myxoid degeneration is a common phenomenon of soft tissue tumors and tumor-like lesions; however, it is rare in cases of esophageal leiomyoma.

Treatment for esophageal tumors includes thoracotomy with esophagectomy, enucleation, and/or tumor resection [1-3]. Uniportal Video-Assisted Thoracic Surgery (VATS) has previously been used to treat esophageal carcinoma in order to reduce surgical trauma [4], and it has been shown to be a feasible treatment for benign disease.

Herein, we report on a case of giant myxoid leiomyoma of the upper esophagus presenting as a giant posterior mediastinal tumor on Computed Tomography (CT) scans. Enucleation of the tumor and primary suture of the esophagus was conducted by uniportal VATS without event.

Case Presentation

A 61-year-old woman with severe dysphagia and dyspnea for about 3 months was admitted to our hospital. Enhanced chest CT showed giant and irregular masses (maximum diameter of 11 cm) in the right posterior aspect of the mediastinum. The masses that were located posteriorly regarding the bronchus and anteriorly regarding the esophagus pulled the superior vena cava and the bronchus (Figures 1A-1D). Esophagoscopy revealed a narrowed esophageal lumen and swollen and eroded lesions of the esophageal mucosa, 18 cm to 24 cm from the incisor (Figure 1E). Bronchoscopy showed an external pressure-induced bulge in the middle and lower part of the trachea, accounting for 50% of the lumen of the trachea (Figure 1F). Narrowing of the esophagus and trachea resulted in

OPEN ACCESS

*Correspondence:

Xiaojun Yao, Department of Thoracic Surgery, The Public Health Clinical Center of Chengdu, No. 18, Jingjusi Road, Jinjiang District, Chengdu 610061, China, Tel: 86 28 84537477; Fax: 86 28 84537477;

E-mail: flyingyao@163.com

Received Date: 20 Oct 2022

Accepted Date: 07 Nov 2022

Published Date: 10 Nov 2022

Citation:

Yao X, Li G, Song Y, Jiang L. Uniportal Video-Assisted Thoracic Surgical Enucleation of a Giant Upper Esophageal Myxoid Leiomyoma: A Case Report. *Clin Surg*. 2022; 7: 3588.

Copyright © 2022 Yao X. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

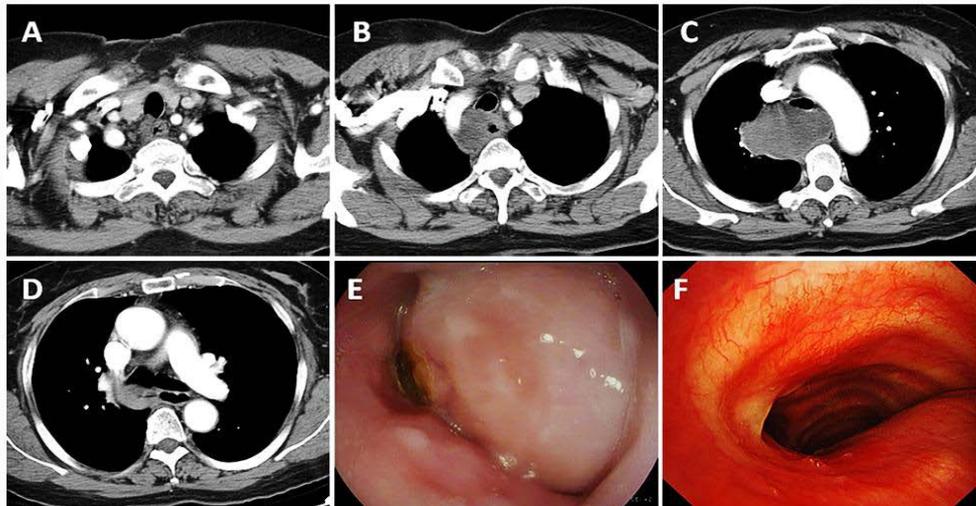


Figure 1: (A-D) Chest CT scan showing an irregular mass in the right posterior upper mediastinum. The mass is close to the right brachiocephalic vein, superior vena cava, right upper pulmonary artery, and azygos vein. (E) Esophagoscopy image showing that the esophageal mucosa is swollen and the upper lumen is narrowed. (F) Bronchoscopy images showing an external pressure-induced bulge in the middle and lower part of the trachea and the opening of the right main bronchus.

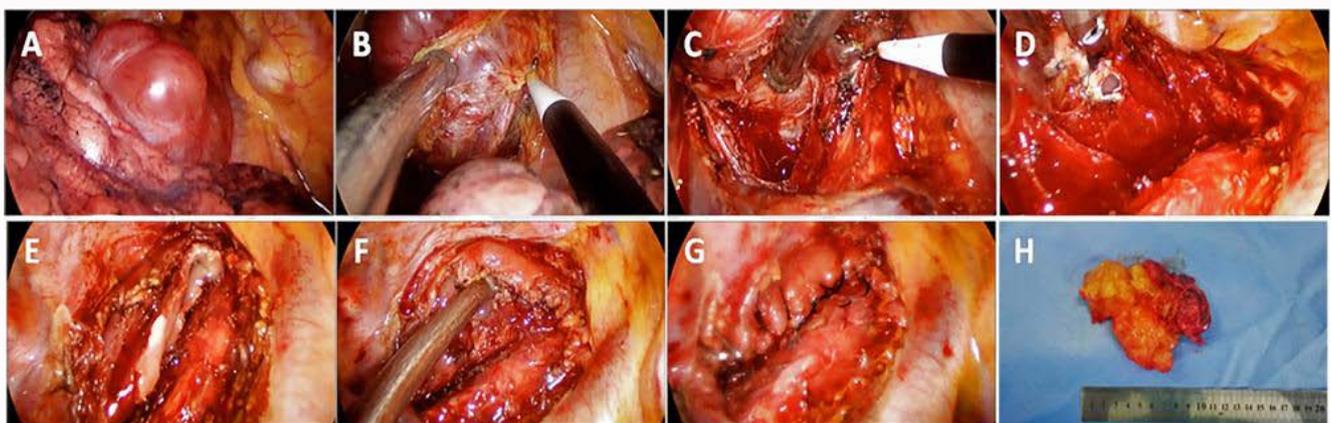


Figure 2: (A-G) The tumor in the right posterior upper mediastinum originated from the lamina propria of the upper thoracic segment of the esophagus. Radical resection was achieved by uniportal VATS. The mucosal defects were continuously sutured with 3-0 antibacterial Vicryl and the muscle layer was intermittently sutured with 3-0 silk thread. (H) Postoperative specimen inspection revealed that the cystic-solid tumor was yellowish white with clear mucus.

dysphagia and dyspnea. Endoscopic Ultrasonography (EUS) was not utilized for the tumor in the thoracic inlet and Endoscopic Bronchial Ultrasonography (EBUS) was not utilized because the patient could not tolerate a long procedure due to dyspnea. Positron Emission Tomography (PET)-CT was not performed because of economic issues.

Uniportal VATS resection of the tumor was performed in order to cure the disease and resolve the serious symptoms, with esophagectomy being found to be unnecessary. Gastrointestinal tract cleaning (sodium phosphate of 75 ml) was performed as the gastrointestinal tract affected during the surgery.

After general anesthesia was initiated, double-lumen endotracheal intubation was utilized to ensure single-lung ventilation. The patient was placed in the left lateral position to explore both the trachea and the esophagus, and a 4-cm incision was made at the 4th intercostal space of the middle axillary line of the right chest wall. The tumor in the right posterior upper mediastinum was found to originate from the lamina propria of the upper segment of the esophagus. The tumor

had a size of 11 cm × 8 cm × 6 cm, with irregular shape and clear boundaries, bulging into the right mediastinum and compressing the trachea, superior vena cava, azygos arch and vagus nerve (Figure 2H). Complete tumor enucleation was achieved after removal of adhesions and partial resection of the esophageal mucosa, which had been invaded by the tumor. A 5 cm long mucosal defect was observed after completion of the enucleation. The esophageal mucosal defect was continuously sutured with 3-0 antibacterial Vicryl and the muscle layer was intermittently sutured with 3-0 silk thread (Figures 2A-2G). No malignant tumor cells were observed in the frozen section examination during the operation.

The postoperative pathological results revealed that the cystic-solid tumor was a yellowish white soft tissue spindle cell tumor with clear mucus (Figure 2F) and myxoid degeneration. It had no necrosis or *heterocyst's* and slight nuclear mitosis (<2/10 high power fields) (Figure 3A, 3B). Immunohistochemistry indicated Vim (+), Des (+), SMA (+), STAT-6 (+), β-Catenin (+), CD34 (-), S-100 (-), SOX-10 (-), PCK (-), and Ki-67 <5% (+) (Figures 3C-3L). The diagnosis of myxoid

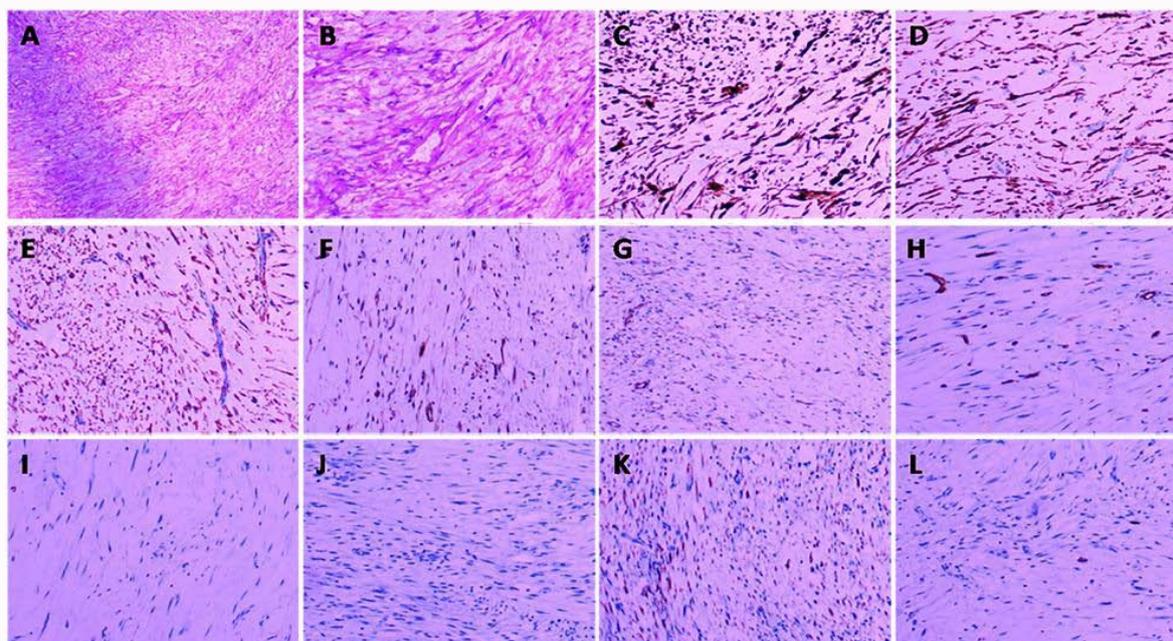


Figure 3: (A) Hematoxylin and Eosin (HE) staining, x400. (B) HE staining, x40. (C) Vim (+). (D) Des (+). (E) SMA (+). (F) STAT-6 (+). (G) β -Catenin (+). (H) CD34 (-). (I) S-100 (-). (J) SOX-10 (-). (K) PCK (-). (L) Ki-67<5% (+).

leiomyoma was confirmed by the immunohistochemistry.

The patient recovered uneventfully and was discharged 7 days after the operation. No stricture of the esophagus was observed during the follow-up period. Esophagoscopy at 6 months after the operation revealed longitudinal scars in the esophagus at 18 cm to 24 cm from the incisor.

Discussion

Leiomyoma is the most common type of benign disease of the esophagus, though it often occurs in the lower and/or gastroesophageal junction [1,5,6]. Leiomyoma in the middle or upper esophagus is rare [7-9]. In our rare case, the esophageal leiomyoma in the upper mediastinum compressed the esophagus and trachea and had a size of 11 cm \times 8 cm \times 6 cm. The esophageal mucosa overlying the esophageal leiomyoma was swollen and eroded, so malignant disease could not initially be excluded. As EBUS and EUS were not suitable for the patient, surgical biopsy was warranted.

The symptoms of esophageal leiomyoma vary according to the degree of the obstruction of the esophagus and/or trachea lumen. Some patient's area symptomatic or experience chest discomfort as the obstruction of the esophagus and/or trachea is slight [10]. Others experience nausea, vomiting, and dyspnea due to serious obstruction of the trachea, while others experience dysphagia due to serious obstruction of the esophagus [11-13]. In our study, the patient had both dyspnea and dysphagia as both the trachea and esophagus were compressed by the giant esophageal leiomyoma, which presented as a huge mediastinal mass on enhanced-CT scans.

Esophagectomy has been utilized as a routine procedure for patients with giant esophageal leiomyoma, even though there are arguments that this is a radical treatment as the disease is benign [14,15]. Deciding between extra-mucosal enucleation and esophagectomy for giant esophageal leiomyoma is challenging [9]. There is a lack of detailed criteria to distinguish the patients that would

benefit from extra-mucosal enucleation rather than esophagectomy. There are no initially observable differences between cases of giant esophageal leiomyoma that are not suitable for simple enucleation and esophageal sarcomas that necessitate esophagectomy [11]. However, for patients with giant esophageal leiomyoma that can be resected without severely damaging the esophageal mucosa, enucleation seems to always be suitable as it causes less trauma and maintains the stability of the digestive system. Derwaiz et al. [16] reported on a series of patients who underwent extra-mucosal enucleation for esophageal leiomyoma, confirming that extra-mucosal enucleation is a safe and feasible option for these patients. A-Lai et al. [17] also reported on the surgical results of extra-mucosal enucleation, demonstrating that thoracotomy is needed when the esophagus is encircled by the leiomyoma. Esophageal mucosal defects represent a challenge regarding extra-mucosal enucleation, as this is difficult to treat postoperatively. Pham et al. [13] reported that intraoperative esophageal mucosal repair avoids postoperative leakage. In our case, an esophageal mucosal defect of about 5 cm was continuously sutured with 3-0 antibacterial Vicryl, and no leakage was observed postoperatively. Closely checking the esophageal mucosa is necessary to observe esophageal defects.

Uniportal-VATS have been used in esophagectomy to reduce postoperative pain [4]. For cases of esophageal leiomyoma, a benign disease, uniportal-VATS is useful. However, it is difficult to enucleate a giant esophageal leiomyoma as the operative view is limited, especially regarding the upper mediastinum. Our patient was placed in the left lateral-position in order for us to explore both the trachea and the esophagus, which could not be done in the semi-prone position. Our case is the first case of upper esophageal leiomyoma enucleated using uniportal-VATS. Robot-assisted thoracoscopic surgery using the da-Vinci surgical system has advantages over VATS in narrow upper mediastinum [7]. But the RATS need more ports to conduct the procedure. Maybe, in the near future, uniportal-RATS could be utilized for this complicated disease with less trauma.

Endoscopic resection has also been proved safe and feasible in the treatment of esophageal leiomyoma [19,20]. But for the leiomyoma located in the thoracic inlet esophagus, endoscopic maybe difficult to treat because of the important surrounding structures. In cases with tumor surrounding the lower esophagus, thoracoscopic surgery combine with endoscopic creation was also safe and feasible [21], which was a potential breakthrough for the treatment of benign esophageal disease.

Using intraoperative frozen section examination, malignancy was excluded, and esophagectomy was avoided. Immunohistochemistry was used to establish the final diagnosis of giant myxoid leiomyoma. The key features of myxoid degeneration are hyaluronic acid and acid mucopolysaccharide [18]. In addition to the myxoid characteristics, primary tumor histological characteristics are found in these tumors.

In summary, we report on the first case of esophageal myxoid leiomyoma presenting as a giant upper mediastinal tumor, which was enucleated by uniportal VATS. Accumulation of such cases is very important for pathologists, radiologists, and surgeons as it can improve the understanding of esophageal myxoid leiomyoma.

Conclusion

In conclusion, giant esophageal myxoid leiomyoma should be considered when a giant upper mediastinal mass is observed on CT scans. Uniportal-VATS may be safe and effective for treating giant upper esophageal myxoid leiomyoma.

Author Contributions

Conceptualization, GL and XJ-Y. Manuscript and literature review, GL and XJ-Y. Image editing, GL, YJ-S and NW. Figures and pathological review, GL and XJ-Y. Manuscript supervision, LS-J and XJ-Y. All authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

References

- Moettinen M, SarlomoRikala M, Sobin LH, Lasota L. Esophageal stromal tumors: A clinicopathologic, immunohistochemical, and molecular genetics study of 17 cases and comparison with esophageal leiomyomas and leiomyosarcomas. *Am J Surg Pathol.* 2000;24(2):211-22.
- Milito P, Asti E, Aiofi A, Zanghi S, Siboni S, Bonarina L. Clinical outcomes of minimally invasive enucleation of leiomyoma of the esophagus and esophagogastric junction. *J Gastrointest Surg.* 2020;24(3):499-504.
- Le DN, Nguyen VH, Duong TN, Than TV. Complete thoracoscopic enucleation of the esophageal leiomyoma at the level of the azygos vein: A case report. *Int J Surg Case Rep.* 2021;88:106537.
- Zhang ZH, Rong BL, Guo MF. Uniportal thoracoscopic McKeown esophagectomy. *Indian J Surg.* 2020;82(4):669-71.
- Sun LJ, Chen X, Dai YN, Xu CF, Ji F, Chen LH, et al. Endoscopic ultrasonography in the diagnosis and treatment strategy choice of esophageal leiomyoma. *Clinics (Sao Paulo).* 2017;72(4):197-201.
- Singla V, Gupta E, Bhattacharjee H, Joshi M, Sharma R, Parshad R. Thoracoscopic enucleation of large esophageal leiomyoma in the lower esophagus: Challenges and solutions. *Indian J Thorac Cardiovasc Surg.* 2021;37(6):694-97.
- Kemuriyama K, Motoyama S, Sato Y, Wakita A, Nagaki Y, Fujita H, et al. Robot-assisted thoracoscopic enucleation for a large esophageal leiomyoma: A case report. *Surg Case Rep.* 2021;7(1):129.
- Hu XX, Lee H. Complete thoracoscopic enucleation of giant leiomyoma of the esophagus: A case report and review of the literature. *J Cardiothorac Surg.* 2014;9:34.
- Elbawab H, Alotaibi AF, Binammar AA, Boumarah DN, Alharbi TM, Alreshaid FT, et al. Giant esophageal leiomyoma: Diagnostic and therapeutic challenges. *Am J Case Rep.* 2021;22:e934557.
- Tang XY, Tan YY, Lv L, Liu DL. Submucosal tunneling endoscopic resection for seven esophageal leiomyomas. *Endoscopy.* 2018;50(3):e59-60.
- Shang QX, Yang YS, Wan WP, Hu WP, Chen LQ. Missed diagnosis of esophageal leiomyoma leading to esophagectomy: A case report and review of literatures. *J Thorac Dis.* 2018;10(1):E65-9.
- Wong T, Pattarapuntakul T, Keeratichananont S, Cattapan K, Nirattisaikul S, Wetwittayakhlung P. Multiple esophageal leiomyoma presenting with clinical dysphagia from mechanical obstruction and motility disorder. *Case Rep Gastroenterol.* 2021;15(3):861-68.
- Pham DH, Nguyen ND, Do ML, Nguyen XH, Quach VK, Bretagnol F, et al. Video assisted thoracoscopy or laparoscopy for enucleation of esophageal leiomyoma: A seven-year single center experience of 75 cases. *J Vis Surg.* 2022;159(2):108-13.
- Cheng BC, Chang S, Mao ZF, Li MJ, Huang J, Wang ZW, et al. Surgical treatment of giant esophageal leiomyoma. *World J Gastroenterol.* 2005;11(27):4258-60.
- Tsai SJ, Lin CC, Chang CW, Hung CY, Shieh TY, Wang HY, et al. Benign esophageal lesions: Endoscopic and pathologic features. *World J Gastroenterol.* 2015;21(4):1091-8.
- Derwaiz A, Shaw VK, Singh A, Chaudhary A. Extra-mucosal enucleation is still a safe and feasible treatment option of giant esophageal leiomyoma. *Indian J Gastroenterol.* 2018;37(1):63-6.
- A-Lai GH, Hu JR, Yao P, Lin YD. Surgical treatment for esophageal leiomyoma: 13 years of experience in a high-volume tertiary hospital. *Front Oncol.* 2022;4(12):876277.
- Zhao T, Liu XS, Lu Y. Myxoid epithelial leiomyoma of the vulva: A case report and literature review. *Case Rep Obstet Gynecol.* 2015;2015:894830.
- Tu SF, Huang SL, Li GH, Tang XW, Qing HT, Gao QP, et al. Submucosal tunnel endoscopic resection for esophageal submucosal tumors: A multicenter study. *Gastroenterol Res Pract.* 2018;2018:2149564.
- Tang Y, Deng XJ, Yang WX, Xiao X. Submucosal tunnel endoscopic resection for a large leiomyoma originating from the muscularis propria in the upper esophagus. *Rev Esp Enferm Dig.* 2022;114(6):348-9.
- Oyama K, Ohuchida K, Shindo K, Moriyama T, Hata Y, Wada M, et al. Thoracoscopic surgery combined with endoscopic creation of a submucosal tunnel for a large complicated esophageal leiomyoma. *Surg Case Rep.* 2022;6(1):92.