



A novel hybrid approach for enucleation of esophageal leiomyoma

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Introduction

Esophageal leiomyoma is the most common benign tumor of the esophagus, which usually arises from the muscularis propria or muscularis mucosae. Incidence of benign esophageal neoplasms was reported to be in the range of 0.005% to 7.9% in autopsy cases, while leiomyomas account for 70–80% of them (1). Options for management include surveillance for small-sized tumors and surgical or endoscopic resection for large or symptomatic ones. Leiomyoma is commonly located in the lower two-thirds of the esophagus. In open surgery, right-sided surgical approach is preferred for lesions in the middle and upper esophagus, and left-sided approach for those in the distal lower esophagus. Video-assisted thoracoscopic surgery (VATS) for esophageal leiomyoma was first described by Everitt in 1992 (2,3). As scattered researches reported, enucleation of benign esophageal tumors can be performed effectively and safely with low morbidity (4-6), mostly through right-sided approach. The major complication after the enucleation of esophageal leiomyomas is mucosal injury, which may lead to esophageal leakage or perforation. We hereby report a novel approach using combined thoracoscopy and endoscopy to facilitate tumor removal and prevent mucosal injury.

Clinical vignette

A 36-year-old man presented to the Shanghai Chest Hospital with complaint of recent onset intermittent dry cough. He was evaluated initially with chest high-resolution computed tomography (HRCT) which showed an irregular shaped mass in the posterior mediastinum measuring 40 mm × 21 mm, compressing the left main bronchus (Figure 1A). Chest magnetic resonance imaging (MRI) with

contrast showed isointensity on T1 and T2 images, with moderate enhancement (Figure 1B). Esophagoscopy showed a smooth submucosal elevated lesion located at 25–32 cm from the incisors, measuring 7 cm in length (Figure 1C). Endoscopic ultrasonography (EUS) showed the tumor was mix and medium echoic predominantly, arising from the muscularis propria (Figure 1D). Esophagogram showed narrowing of the esophageal lumen but with a smooth lining of mucosa (Figure 2A). The patient also had a past history of endoscopic biopsy from the lesion 4 years ago with a diagnosis of leiomyoma. On the basis of the medical history, previous biopsy and imaging, he was diagnosed of having esophageal leiomyoma. A hybrid approach with endoscopy and thoracoscopy was performed to remove the lesion completely without mucosal injury. The final histopathological examination confirmed the diagnosis as esophageal leiomyoma, measuring 70 mm × 30 mm × 28 mm (Figure 2B).

The postoperative course was uneventful. The patient was required to remain fasting only on the operative day. Chest X-ray was normal on the first postoperative day (POD). Esophagogram showed an adequate transit with no leakage or narrowing of the esophagus. Nasogastric tube was removed right afterwards and oral liquid resumed on the same day, which proceeded to semisolid diet on POD2. The drainage tubes were removed on POD2 and POD3. The patient recovered uneventfully and was discharged on POD4.

Operative techniques

The operation was carried out under general anesthesia with double-lumen intubation. The patient was placed in left decubitus position and the right lung was completely

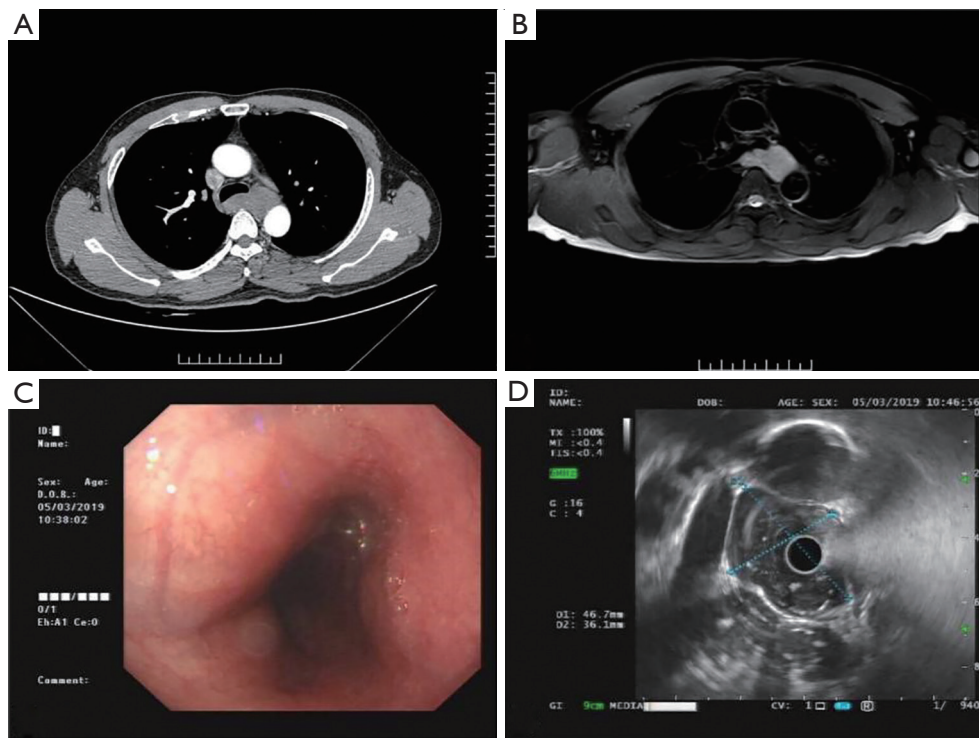


Figure 1 Manifestations of the preoperative examinations.

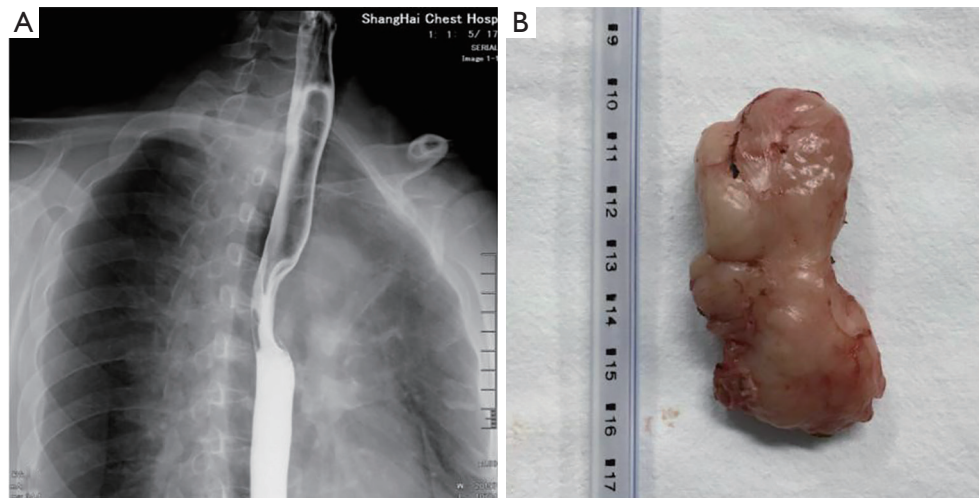


Figure 2 Esophagogram and the specimen of the tumor.

collapsed. A three-port video-assisted thoracoscopic procedure was performed. A 30-degree thoracoscope was positioned through the camera port, located in the seventh intercostal space at the middle axillary line. The other two ports included a working port in the 4th intercostal space at the anterior axillary line and an auxiliary port in the 6th

intercostal space at the posterior axillary line. The right lung was retracted towards the anterior mediastinum to expose the esophagus. The mass was detected in the upper posterior mediastinum above the level of azygous arch. The mediastinal pleura was opened longitudinally after the lesion was found, and a longitudinal myotomy was performed over

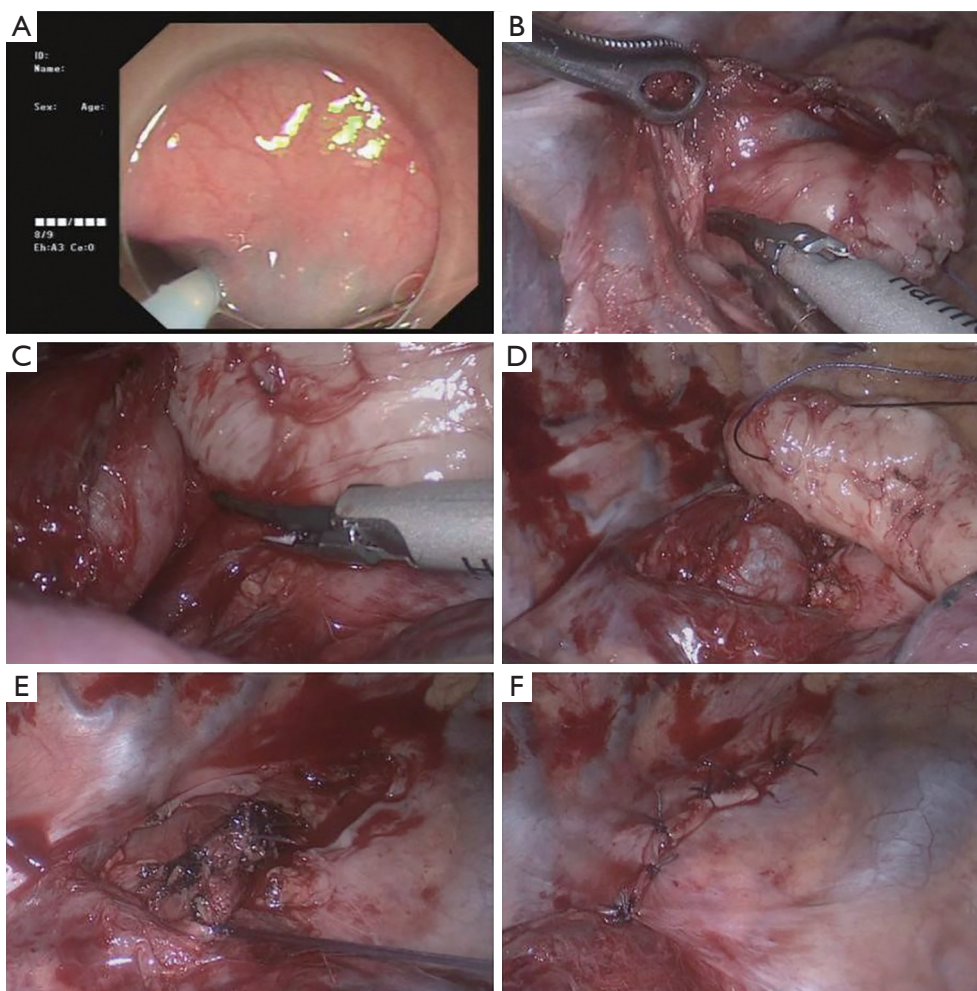


Figure 3 The process of video-assisted thoracoscopic enucleation of the tumor.

the lesion. At the same time, the lesion was also located under esophagoscopy. A methylene blue solution was injected into the submucosal layer under esophagoscopy to make the mucosa around the lesion protruded, blue-stained and separated from the muscular layer, so that the interface between mucosa and the tumor could be easily delineated under thoracoscopy so as to avoid the inadvertent mucosal injury (*Figure 3A*). The tumor was then dissected away from the mucosa gently (*Figure 3B,C,D*), with the help of a traction suture placed in the mass (*Figure 3D*). The resected tumor was taken out through the working port. Air inflation of the esophagus was done under esophagoscopy to confirm the integrity of the mucosa and no air leakage was detected. Subsequently, interrupted absorbable sutures were used to close the muscular layer and then the mediastinal pleura longitudinally (*Figure 3E,F*). Finally,

after careful hemostasis, a 28-Fr chest drain tube was placed through the camera port for postoperative drainage and a small negative suction tube was placed along the incision in esophagus. Operation time was within an hour and total blood loss was less than 100 mL (*Figure 4*).

Comments

Leiomyoma is the commonest benign esophageal tumor, which is usually asymptomatic and may be discovered incidentally. It can be diagnosed on esophagogram, endoscopy, EUS or other examinations like contrast enhanced computed tomography scan and MRI. Endoscopic biopsy may be usually avoided as it may cause the inflammatory adhesion and increasing operative difficulty and complications potentially (8,9). It is suggested that

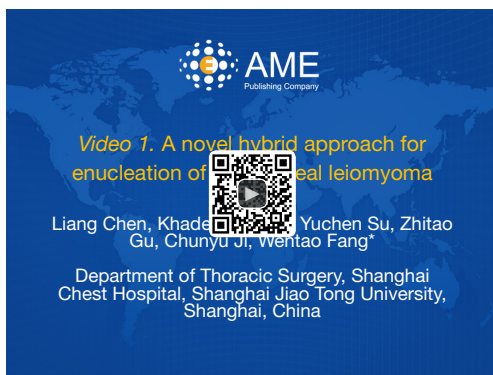


Figure 4 A novel hybrid approach for enucleation of esophageal leiomyoma (7).

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surgery is indicated for tumors larger than 1 cm, potentially malignant tumors, and symptomatic patients since the symptoms can be well relieved after the surgery according to a 40-year pathologic database of the Massachusetts General Hospital (10,11). VATS has been widely used in the enucleation of esophageal leiomyoma and has replaced traditional open thoracotomy gradually since first reported by Everitt in 1992 (2). Compared with open thoracotomy, minimally invasive procedure results in less pulmonary complication, limited operative trauma, less postoperative wound-related pain, more rapid recovery, shorter hospital stay and better wound cosmetics (1). Nevertheless, open thoracotomy or even partial esophagectomy may be required in case of relatively large tumor size, irregular shape, difficult location or technical limitation (12).

In general, for tumors within 1 to 5 cm in diameter, thoracoscopic enucleation is strongly recommended. And intraoperative endoscopy has already been used to locate the small tumors from inside the lumen during surgery and to inspect the mucosal integrity after enucleation (8,13). For tumors circumfusing the esophagus, transillumination under endoscopy can help to identify anatomic landmarks (14). As a new technique, submucosal tunneling endoscopic resection (STER) is also used in endoscopic resection of submucosal tumors (SMTs) in the esophagus with an entry to the submucosal space from the longitudinal mucosal incision. But it is associated with relatively high risks of adverse events and technical difficulties when the tumors are larger than 3 cm in diameter or of irregular shape (15). Thoracoscopic enucleation of leiomyoma of esophagus with the assistance of a balloon-mounted intraluminal endoscope, another new surgical approach, was also

reported previously to reduce the difficulty in grasping the tumor and the risk of mucosal injury (16,17). However, it is usually difficult to identify the border between the tumor and the mucosa when the tumor is larger than 5 cm or irregular-shaped. And dense adhesion of mucosa to the tumor caused by previous biopsy or repeated inflammation may further increase the difficulty in surgical dissection and the risk of mucosal perforation during surgery. In these circumstances, incidence of conversion to thoracotomy and mucosal injury is relatively high (11). Even if esophageal leakage does not happen, oral intakes are often postponed for fear of undetected small mucosa perforation.

In the clinical case discussed above, the preoperative examinations showed that the tumor was large-sized and irregular-shaped. And the patient had received endoscopic biopsy before. Hence dissection was expected to be difficult and mucosal injury during the surgery was a great concern. With the help of simultaneous submucosal injection of methylene blue solution under endoscopy, esophageal mucosa was clearly identified and easily dissected away from the tumor. As a result, enucleation of the esophageal leiomyoma was readily achieved under VATS. And esophageal mucosa was well preserved without perforation or injury during operation. The procedure was smooth and uneventful, with limited bleeding and operation time. And the patient recovered very rapidly from the operation. Oral diet could be resumed on the first post-operative day. This novel approach with combined use of VATS and endoscopy has never been reported before. Our initial experience shows that it is safe, feasible, and helpful for enhanced recovery.

Some points should be noted as follows. The indications for performing this combined surgery remain unknown and require further study. And to what extent this combined approach is helpful and more effective than open thoracotomy is to be answered by more experience.

In conclusion, we report the first case of video-assisted thoracoscopic enucleation of esophageal leiomyoma with combined use of mucosal elevation through endoscopic injection. It is helpful in facilitating tumor resection and in preventing mucosal injury for large-sized, irregular-shaped intramural tumors. Clinical studies need to be carried out to further prove the effectiveness of this novel procedure.

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Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The authors are 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. Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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