# Discussion on surgical approaches for giant, well-differentiated liposarcomas of the esophagus: Report of two cases

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**Abstract.** Giant liposarcoma of the esophagus is an exceedingly rare esophageal tumor with complex treatment options. This study investigated the treatment modalities and reported on the clinical outcomes of two cases involving giant, well-differentiated liposarcoma of the esophagus, providing a reference for the management of similar cases. Both tumors measured >20 cm in length and had diameters exceeding 4 cm; one case exhibited a lobulated appearance with visibly expanded blood vessels on its surface. Following discussions within a multidisciplinary team, under general anesthesia with tracheal intubation, the endoscopy team conducted endoscopic submucosal dissection, collaborating with the thoracic surgery team to manage potential bleeding risks. Both patients successfully underwent endoscopic tumor removal with postoperative pathology confirming the presence of well-differentiated liposarcoma and no observed complications. For patients with giant and complex well-differentiated liposarcomas, endoscopic dissection in conjunction with multidisciplinary collaboration represents a safe and effective treatment option, ensuring complete tumor removal while minimizing surgical trauma and enhancing patient prognosis.

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*Abbreviations:* CT, computed tomography; ESD, endoscopic submucosal dissection; FISH, fluorescence *in situ* hybridization; P16, cyclin-dependent kinase inhibitor 2A; Vim, Vimentin; CK4, cytokeratin 4; MDM2, mouse double minute 2 homolog; S100, S100 calcium-binding protein; Sox10, SRY-box transcription factor 10; SMA, smooth muscle actin; Melan-A, melanocyte antigen; HMB45, human melanoma black 45; DOG-1, discovered on GIST-1; Ki-67, Ki-67 antigen; P53, tumor protein P53

*Key words:* liposarcoma, esophagus, surgical discussion

## Introduction

Liposarcoma is a rare malignant tumor originating from adipocytes, accounting for 15-20% of soft tissue sarcomas (1). The differences among its subtypes primarily depend on immunohistochemical characteristics, cell morphology, and genetic variations. According to the 2020 World Health Organization Classification of Soft Tissue and Bone Tumors (2), liposarcoma can be divided into well-differentiated liposarcoma (atypical lipomatous tumor), dedifferentiated liposarcoma, myxoid liposarcoma, pleomorphic liposarcoma, and myxoid pleomorphic liposarcoma, with well-differentiated liposarcoma being the most common type. Each subtype exhibits significant differences in clinical presentation, biological characteristics, and drug sensitivity (3,4). Esophageal liposarcomas are exceedingly rare, constituting only 1.2-1.5% of gastrointestinal liposarcomas (5). Studies indicate that minimally invasive procedures, such as transoral and thoracoscopic techniques, are the preferred surgical methods for treating esophageal liposarcomas. However, challenges such as risk of bleeding and high recurrence rates remain for complex giant esophageal liposarcomas (6,7). Surgical resection is essential for managing giant, well-differentiated liposarcomas of the esophagus; however, no standardized protocol exists for selecting the surgical approach.

Given the rarity of esophageal liposarcomas and the complexity of their treatment, evaluating the effectiveness of different surgical methods holds clinical significance. By discussing the surgical approaches for two cases of giant, well-differentiated esophageal liposarcoma, through multidisciplinary collaboration, leveraging the strengths of both endoscopic techniques and surgical interventions, complete tumor excision was achieved and both patients demonstrated favorable postoperative recovery. This article aims to provide additional treatment options and shared experiences to enhance patient prognosis and quality of life.

## Case report

Both patients were admitted due to dysphagia. Patient 1 was a 62-year-old male who reported 'dysphagia for 1 month, which worsened 1 week previously'. One month prior to admission to an external Hospital in May 2024, the patient underwent laryngoscopy and was diagnosed with pharyngitis. The condition showed slight improvement following anti-inflammatory

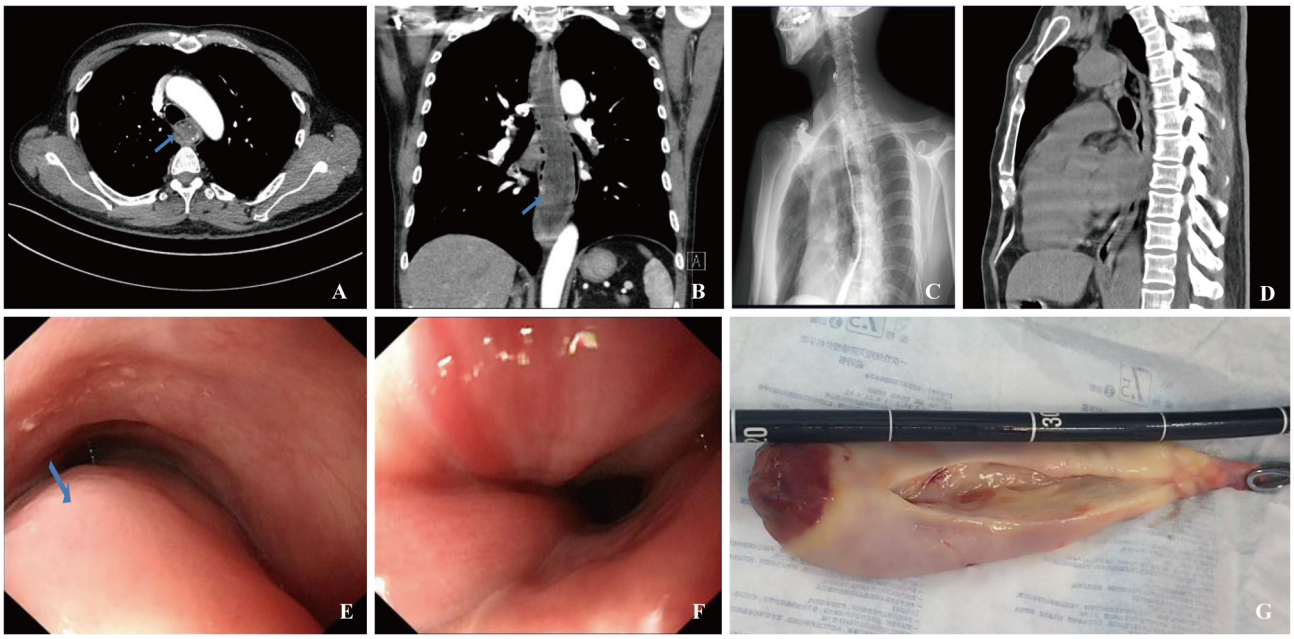


Figure 1. Patient 1: Enhanced chest CT scans in the (A) axial and (B) coronal planes, revealing a large cystic lesion within the esophagus. (C) Upper gastrointestinal imaging (a type of examination that utilizes X-rays and contrast agents) performed on the first postoperative day, showing no extravasation of the contrast agent or filling defects. (D) Follow-up chest CT 1 month after surgery, confirming that the esophagus remains patent. (E) Gastroscopy images show a large elongated mass within the lumen of the esophagus. (F) Gastroscopy images demonstrating significant submucosal elevation of the esophageal mucosa, originating from the base. (G) The gross tumor specimen (each scale in the image represents 5 centimeters). The blue arrow indicates the esophageal tumor.

treatment. However, 1 week before admission to the Affiliated Hospital of Guizhou Medical University (Guiyang, China) in June 2024, the patient's dysphagia worsened, accompanied by difficulty eating. Of note, the patient was able to expel the tumor from the esophagus through the mouth and swallow it back into the body. The patient had a history of varicose vein surgery in both lower limbs and a 10-year smoking history (20 cigarettes per day, quit 1 month ago). Upon admission, physical examination revealed a mass in the esophagus that could partially be vomited out. Chest computed tomography (CT; Fig. 1A and B) revealed esophageal dilation, with a mass-like lesion exhibiting increased cystic density and no obvious enhancement on the enhancement scan. The area near the gastroesophageal junction displayed uneven enhancement, with no fat density components observed. The esophageal tumor measured ~23 cm in length, with its base located in the upper segment of the esophagus, parallel to the level of the T1 vertebra. In addition, multiple lymph node calcifications were visible in both lung hila and the mediastinum. Gastroscopy revealed a giant submucosal lesion with expanded blood vessels and inflammatory exudate (Fig. 1E and F).

Patient 2 was a 41-year-old male who presented to the Affiliated Hospital of Guizhou Medical University (Guiyang, China) in August 2024 due to 'dysphagia and recurrent fever for 1 month'. The patient experienced swallowing discomfort of unknown causes, with a maximum body temperature of 38.8˚C. Self-medication with anti-inflammatory drugs provided no significant improvement and the patient had lost 4 kg in weight over the past month. The patient had a 20-year smoking history (20 cigarettes per day) and occasionally consumed alcohol, with no other significant health issues. Physical examination revealed no remarkable findings. Enhanced chest CT (Fig. 2A and B) revealed a large strip-like lesion in the esophagus, exhibiting mixed density comprising both solid and fatty components. The lesion measured 3.1x6.1x20.2 cm. The mediastinal lymph nodes appeared normal and the CT also demonstrated prominent vessels surrounding the tumor. Endoscopic examination (Fig. 2F and G) provided a more direct and clearer view, revealing a strip-like elevation from the upper segment of the esophagus (16 cm from the incisors) to the lower segment (35 cm from the incisors). The upper segment lesion showed congestion and superficial ulceration, with localized gray-blue blood vessels. Endoscopic ultrasound examination indicated that the elevation originated from the third layer and presented as a mixed echogenic mass (Fig. 2E). In order to clarify the diagnosis before surgery, two patients underwent a biopsy. The biopsy results of Patient 1 showed inflammatory exudate and necrosis (Fig. 3A); the biopsy results of Patient 2 exhibited moderate chronic inflammation, erosion, and lymphoid tissue hyperplasia forming follicles (Fig. 4A). Both were initially diagnosed as benign esophageal tumors.

In the surgical management, both patients were considered to have benign esophageal tumors located in the mucosal layer. After discussions among experts from the endoscopy center and thoracic surgery, endoscopic tumor removal was prioritized. In case the endoscopic procedure could not be completed, the thoracic surgery team was prepared to perform open resection via a cervical approach. Both patients ultimately underwent endoscopic tumor removal. Patient 1 underwent surgery on the third day after admission, during which a large, strap-like lesion was found on the left esophageal wall, ~38 cm from the incisors, occupying about two-thirds of the esophagus. The tumor measured 22x4x4 cm (Fig. 1G) and was successfully removed endoscopically. Patient 2 underwent surgery on the fifth day after admission, during which a giant sessile tumor

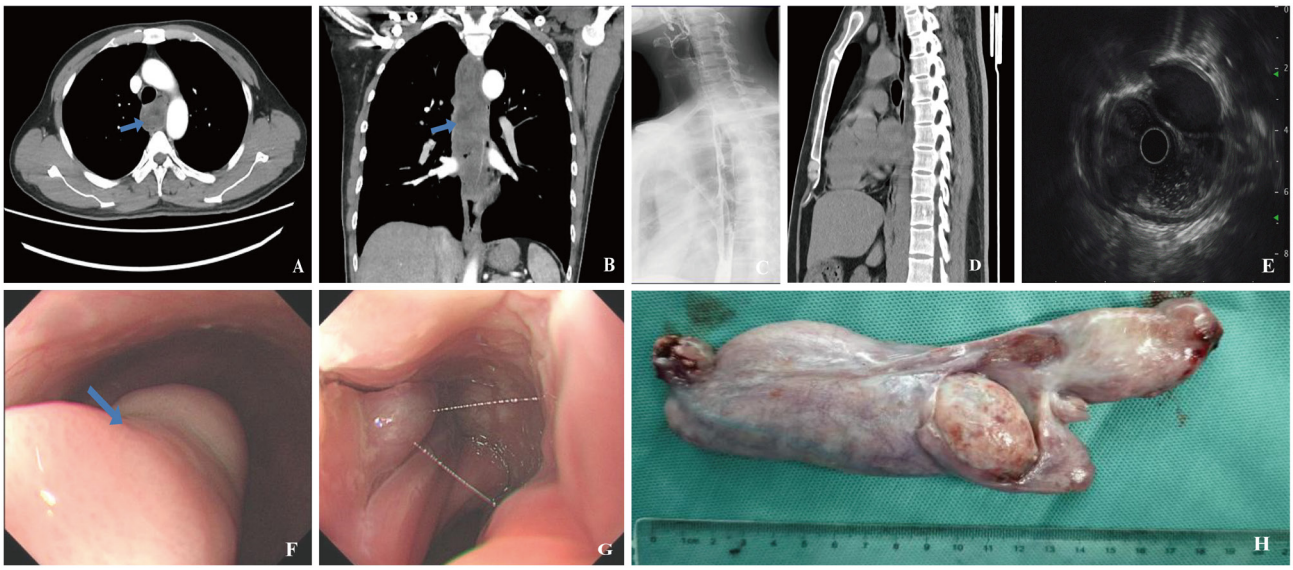


Figure 2. Patient 2: Enhanced chest CT scans in the (A) axial and (B) coronal planes, revealing a large, strip-like, mixed-density shadow within the esophagus, containing solid components with uneven enhancement. (C) Upper gastrointestinal imaging (a type of examination that utilizes X-rays and contrast agents) on the first postoperative day, showing no extravasation of the contrast agent or filling defects. (D) Follow-up chest CT 1 month after surgery, confirming esophageal patency. (E) The endoscopic ultrasound indicates that the lesion originates from the third layer, presenting as a mass with mixed echogenicity and uneven internal echoes. (F) Gastroscopy images indicate a strip-like elevation in the esophagus. (G) Gastroscopy images indicate a tubular tumor accompanied by visible dilated blood vessels and inflammatory exudate. (H) The gross tumor specimen. The blue arrow indicates the esophageal tumor.

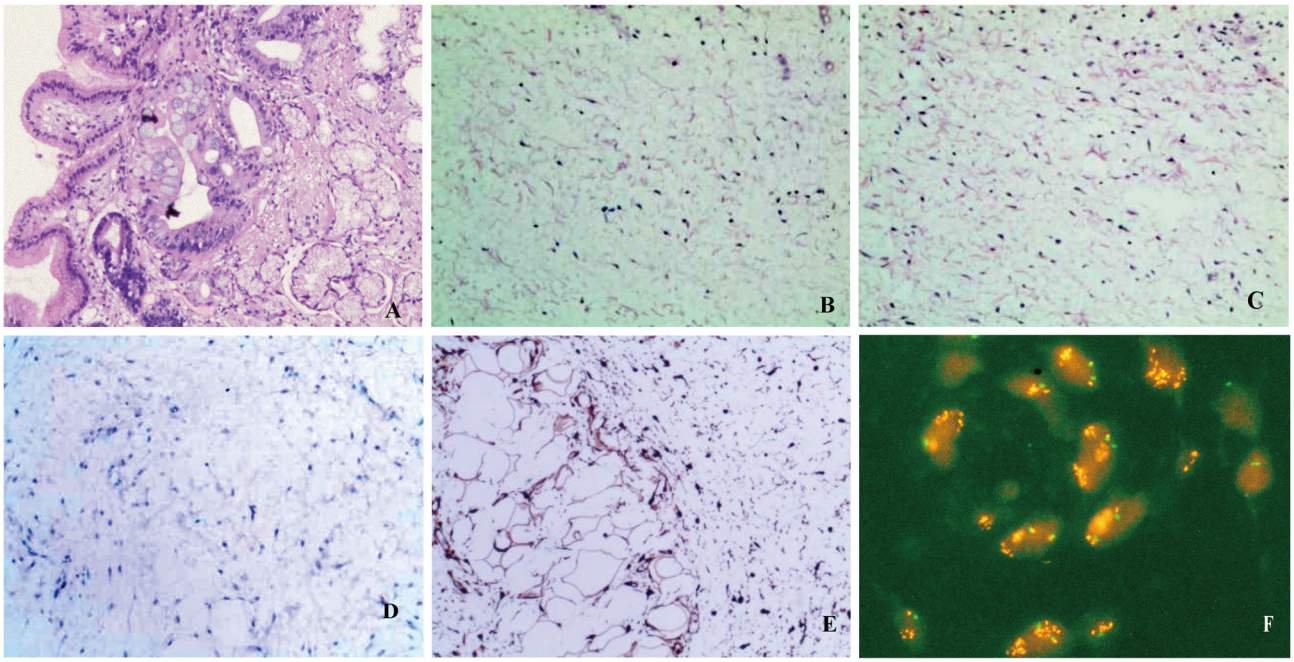


Figure 3. Patient 1: Pathological findings. (A) The results of the gastric biopsy with H&E staining indicate the presence of inflammatory exudates and necrosis (magnification, x200). (B) H&E staining indicated the formation of mucosal ulcers, with spindle-shaped cells and vascular proliferation observed beneath the ulcers (magnification, x100). (C) The results of H&E staining under high-power microscopy showed that spindle-shaped cells are distributed among the adipocytes, with certain areas exhibiting characteristics of mucinous degeneration. (D) Immunohistochemical staining for MDM2, which was negative. (E) Immunohistochemical staining for P16, which was positive. (F) Chromosome fluorescence *in situ* hybridization using an *MDM2* (12q15) gene probe, revealing a positive result for *MDM2* gene amplification (magnification, x200). MDM2, mouse double minute 2 homolog.

was found, extending 16 to 36 cm from the incisors, with its base located in the neck, lobulated and occupying two-thirds of the esophagus. The tumor measured 20x6x3 cm (Fig. 2H) and exhibited a soft consistency, with two prominent blood vessels (~0.4 cm in diameter) visible on the surface.

Both procedures followed a similar approach. Attempts to use a snare and harmonic scalpel were unsuccessful, so hemostatic forceps were employed to coagulate visible blood vessels at the tumor's base. The lesion was dissected using a disposable endoscopic scalpel starting from the mucosa and extending to

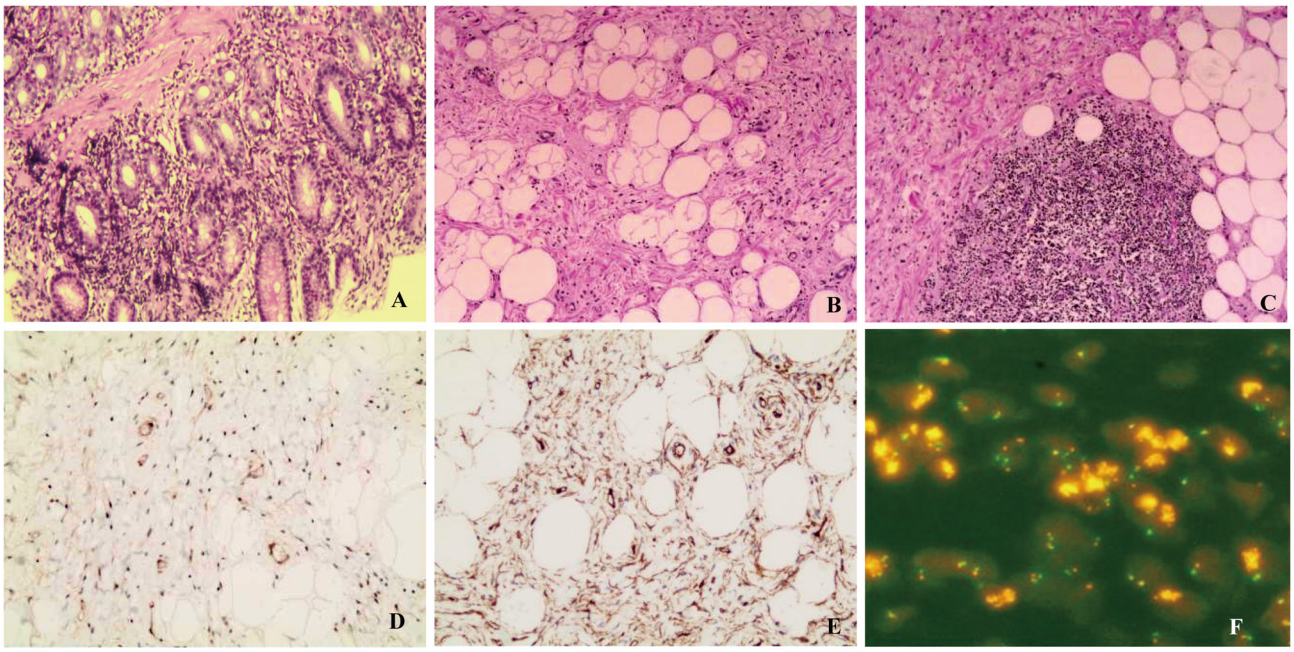


Figure 4. Patient 2: Pathological findings. (A) The results of the endoscopic ultrasound biopsy show: moderate chronic inflammation in the mucosa, accompanied by erosion, as well as lymphoid tissue hyperplasia forming lymphoid follicles. (B) H&E staining demonstrating widespread proliferation of fibrous adipose tissue in the esophageal submucosa, forming fat lobules of varying sizes. (C) H&E staining demonstrating scattered thick-walled blood vessels and focal lymphoid tissue proliferation is observed. (D) Immunohistochemical staining for *MDM2*, showing scattered positive reactions. (E) Immunohistochemical staining for *P16*, which was positive. (F) Chromosome fluorescence *in situ* hybridization using an *MDM2* (12q15) gene probe, revealing positive *MDM2* gene amplification (magnification, x200). MDM2, mouse double minute 2 homolog.

the submucosal layer, revealing white fibrous tissue and large blood vessels. After electrocautery, the tumor was carefully extracted until significant resistance was encountered at the entrance of the esophagus; repeated attempts to change angles only resulted in partial tumor extraction, prompting the use of oval and tongue forceps to drag the tumor out of the oral cavity. Postoperative pathology confirmed well-differentiated liposarcoma of the esophagus. In Patient 1, the esophageal lesion presented as a mucosal ulcer, with spindle-like cells and vascular proliferation observed beneath the ulcer, and certain areas showing mucinous degeneration (Fig. 3B and C). Immunohistochemical results showed negative Cytokeratin (CK), with positive Vimentin (Vim), cyclin-dependent kinase inhibitor 2A (P16; Fig. 3E), CDK4 and CD34 and negative mouse double minute 2 homolog (MDM2; Fig. 3D), SRY-box transcription factor 10 (Sox10), desmin, smooth muscle actin (SMA), caldesmon, melanocyte antigen (Melan-A), human melanoma black 45 (HMB45), CD117 and discovered on GIST-1 (DOG-1); Ki-67 antigen (Ki-67) was ~3% positive. Fluorescence *in situ* hybridization (FISH) (Fig. 3F) analysis indicated positive amplification of the *MDM2* gene (12q15). In Patient 2, there was significant proliferation of fibrofatty tissue in the submucosal layer of the esophageal lesion, forming fat lobules of varying sizes, accompanied by thick-walled blood vessels and focal lymphoid tissue hyperplasia (Fig. 4B and C). Immunohistochemical results showed atypical cells positive for Vim, S100, CD34, desmin, P16 (Fig. 4E), MDM2 (Fig. 4D) and CDK4 and negative for SMA, caldesmon, DOG-1 and CD117; and Ki-67 was ~5% positive (primary antibody details may be viewed in Table SI). FISH analysis (Fig. 4F) indicated positive amplification of the *MDM2* gene (for detailed pathological methods, please refer to Data S1). Following endoscopic surgical resection, neither patient experienced significant discomfort or complications. On the first postoperative day, imaging (Figs. 1C and 2D) confirmed a normal esophageal lumen. A follow-up chest CT 1 month after surgery showed that the esophageal lumen remained unobstructed, suggesting a good prognosis (Figs. 1D and 2E).

## Discussion

The incidence of esophageal liposarcomas is low, with most cases originating from the mucosal layer; mesenchymal tumors account for only 5% (8). The two cases reported in the present study were both well-differentiated liposarcomas, characterized by slow growth and low metastatic potential but with a risk of local recurrence and resistance to chemotherapy and radiotherapy (9). Research indicates that the development of well-differentiated liposarcomas is associated with repeated amplification of chromosome 12, which provides an important direction for targeted therapy (10). Related studies indicate that esophageal liposarcomas predominantly occur in males, with an average onset age of 58.4 years (range, 38-73 years). The tumor typically extends from the cervical segment to the thoracic segment. However, its nonspecific clinical manifestations pose challenges for accurate diagnosis (11-14).

In the present study, in the context of esophageal liposarcoma, preoperative chest enhanced CT and esophagogastroscopy were particularly important. These two diagnostic methods not only provided a clear delineation of the tumor and surrounding structures but also effectively assessed the tumors' vascular supply. Chest enhanced CT has a unique

advantage in visualizing the vessels around the tumor; the use of contrast agents allows for effective differentiation between the tumor and adjacent tissues, as well as clarifying the blood supply status at the tumor margins. This imaging technique provided crucial information for evaluating the tumors' vascularity. By contrast, endoscopic examination offers a more direct and clearer observational perspective. Through endoscopy, physicians can view the detailed structures of the tumor and its surrounding vessels in real-time, enabling direct assessment and, when necessary, performing biopsies or other interventions to obtain comprehensive diagnostic information. Typically, preoperative endoscopy reveals a pedunculated tumor that could easily be mistaken for a benign lesion, with a definitive diagnosis relying on postoperative pathological analysis. The occurrence of high-grade dedifferentiated liposarcoma is closely related to the amplification of the 12q13-15 chromosomal region, while the immunohistochemical triad of MDM2, CDK4 and p16 serves as its core diagnostic markers (15). *MDM2* gene amplification testing is considered the most reliable method for distinguishing well-differentiated liposarcomas from benign lipomas (16).

Currently, there is no standardized treatment protocol for esophageal liposarcomas and surgical resection remains the primary treatment. Achieving a negative margin during complete resection is essential for a curative outcome and for effectively relieving tumor-induced obstruction. Surgical approaches vary and include total esophagectomy, subtotal esophagectomy and minimally invasive techniques. The choice of surgical approach typically depends on tumor location and size, with options including transcervical, thoracoscopic or endoscopic access routes. Historically, open surgery has been the gold standard (6,17-19). However, with advancements in minimally invasive techniques, these procedures have become the preferred treatment due to their benefits, including reduced postoperative pain, shorter hospital stays and faster recovery. Endoscopic submucosal dissection (ESD) has emerged as a promising, less invasive alternative, particularly for smaller tumors with a stalk and no involvement of large blood vessels. A recent study reported successful endoscopic resection of well-differentiated esophageal liposarcomas, including a tumor measuring 8.3x4.2x2.3 cm, demonstrating the feasibility of ESD (20). In cases where the tumor volume is large and complete endoscopic resection is not possible, or if large blood vessels are involved, cervical incision for resection is a viable option.

The present study reported two cases of giant esophageal tumors, each exceeding 20 cm in length and 4 cm in diameter. Gastroscopy findings revealed visible expanded blood vessels on the surface, with one tumor exhibiting a lobulated appearance, complicating endoscopic removal. After multidisciplinary discussions among thoracic surgery and endoscopy experts, both tumors were presumed to be benign based on gastroscopy and endoscopic ultrasound results. As their stalks were located in the cervical region and confined to the mucosal layer without muscular invasion, endoscopic resection was considered feasible. However, given the large tumor sizes and vascular involvement, the risk of bleeding was high, and limited working space during endoscopy increased the potential for incomplete resection and recurrence. Therefore, thoracic surgery support was necessary to manage potential complications. Following informed consent from the patients and their families, a multidisciplinary surgical approach was adopted. Under general anesthesia with tracheal intubation, the endoscopy team performed ESD, while the thoracic surgery team provided standby support for rapid intervention in case of significant bleeding or other complications beyond endoscopic management. This collaborative approach ensured patient safety while minimizing surgical trauma. Ultimately, both tumors were successfully removed endoscopically without complications and both patients had favorable postoperative outcomes without requiring any lymph node clearance or adjuvant therapy.

The insights from this report are as follows: When patients present with swallowing difficulties, a chest CT scan should be considered, and if necessary, endoscopy should be performed to prevent excessive tumor growth and associated treatment challenges. Prior to endoscopic resection of esophageal liposarcomas, it is crucial to determine tumor malignancy and stalk location to determine whether esophageal resection is necessary. Ensuring that the tumor is confined to the mucosal layer without muscular invasion helps minimize the risk of esophageal perforation (21). For large tumors or cases with significant vascular involvement, procedures should be conducted under general anesthesia in an operating room equipped for emergency open surgery if needed in order to handle unexpected situations and ensure surgical safety. The surgeon should have extensive experience and blood vessels should be pre-coagulated to minimize intraoperative bleeding during tumor resection. Superficial excision should be avoided to prevent recurrence, while overly deep excision could increase the risk of esophageal perforation (22). Large tumors may be difficult to extract through the oropharynx, so it is recommended to use sterile surgical forceps guided by an endoscope to grasp the tumor through the oral cavity, ensuring the safety and effectiveness of the procedure. Postoperatively, patients should refrain from eating for 3 days and receive jejunal or parenteral nutrition (23). Follow-up imaging, including chest CT or upper gastrointestinal studies, should be conducted promptly to monitor for esophageal fistulas. A follow-up chest CT or endoscopy is recommended 6 months to 1 year post-surgery to detect any potential local recurrence (24).

In conclusion, for giant and complex well-differentiated liposarcomas of the esophagus, endoscopic surgery combined with multidisciplinary collaboration is a safe and effective treatment strategy. This approach enables complete tumor removal while minimizing surgical trauma and ensuring favorable patient outcomes. Future studies should continue to accumulate case data to refine treatment strategies, improve endoscopic surgery success rates and strengthen clinical evidence. In addition, further exploration of minimally invasive techniques in esophageal tumor management will be crucial for enhancing patient prognosis and quality of life.

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## Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

## Authors' contributions

JM was responsible for the conceptualization of the study and wrote the manuscript. LL collected data and organized images. XD participated in the conception and design of the study, was responsible for data analysis and interpretation and guided the revision of the content. JM, LL and XD confirm the authenticity of all the raw data. All authors have read and approved the final version of the manuscript..

## Ethics approval and consent to participate

Not applicable.

## Patient consent for publication

Both patients have provided written informed consent regarding the publication of this case report and related images.

## Competing interests

The authors declare that they have no competing interests.

## Use of artificial intelligence tools

During the preparation of this work, AI tools were used to improve the readability, and subsequently, the authors revised and edited the content produced by the AI tools as necessary, taking full responsibility for the ultimate content of the present manuscript.

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