

Recurrent giant esophageal liposarcoma that was successfully treated by surgery: A case report



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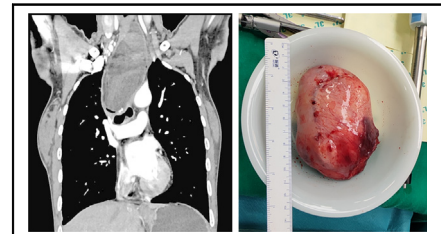
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Location of tumor in the upper part of esophagus and the encapsulated tumor after surgery.

CENTRAL MESSAGE

A strategy for recurrent giant esophageal liposarcoma treatment is discussed.

In March 2018, a 43-year-old female patient received her initial diagnosis of esophageal liposarcoma. Subsequently, she underwent a tumor enucleation through a minimally invasive surgery by thoracoscopy. Pathologic testing confirmed the presence of a well-differentiated liposarcoma and negative margins. However, in November 2022, the patient presented with dysphagia, leading to the diagnosis of tumor recurrence. Despite the recurrence, the patient opted against further treatment. As the tumor continued to progress, the patient's condition worsened, culminating in sudden dyspnea in April 2023. A computed tomography scan revealed a severe tracheal obstruction, the result of compression by a substantial mass measuring $6.3 \times 4.6 \times 11.4$ cm. The tumor was situated in the cervicothoracic junction (Figure 1). Further esophagoscopy confirmed the presence of an esophageal tumor with the mucosal layer intact.

Our multidisciplinary team collectively decided to pursue an esophagectomy. On April 25, 2023, the patient underwent an esophagectomy and tracheotomy (3-incision approach). At the outset, conventional intubation was employed for chest and abdominal surgery. However, due to the prolonged compression of the trachea by the tumor and the significant risk of tracheal malacia, a subsequent tracheotomy was performed following the anastomosis in the neck. The surgical procedure proved to be particularly challenging due to the patient's extensive intrathoracic adhesions and the considerable size of the tumor. Moreover,

the tumor exhibited local invasion into the superior vena cava, further complicating the procedure (Figure 2). Fortunately, the surgery was successfully carried out. The specimen showed the tumor was intraluminal and the results of the pathologic examination and subsequent immunohistochemistry tests, coupled with the detection of MDM2 amplification, confirmed the diagnosis of a well-differentiated liposarcoma. The patient experienced a successful recovery and was discharged from the hospital 1 month after the surgery (Figure E1). The patient provided informed written consent for the publication of this study (institutional review board [2018]273, October 26, 2018).

DISCUSSION

Esophageal liposarcoma is an exceedingly uncommon disease, with its initial documentation attributed to Mansour and colleagues¹ in 1983. Surgical intervention remains the primary therapeutic approach, mainly involving tumor enucleation.² However, the intricacies associated with managing the condition post-tumor recurrence are consistently challenging.

In this case, the unusual recurrence of esophageal liposarcoma has precipitated a critical medical situation for the patient. Given the tumor's considerable size and the potential risks associated with a repeat operation, our initial exploration has focused on seeking alternative treatment avenues. However, whether neoadjuvant chemotherapy and radiotherapy can effectively induce tumor shrinkage remains

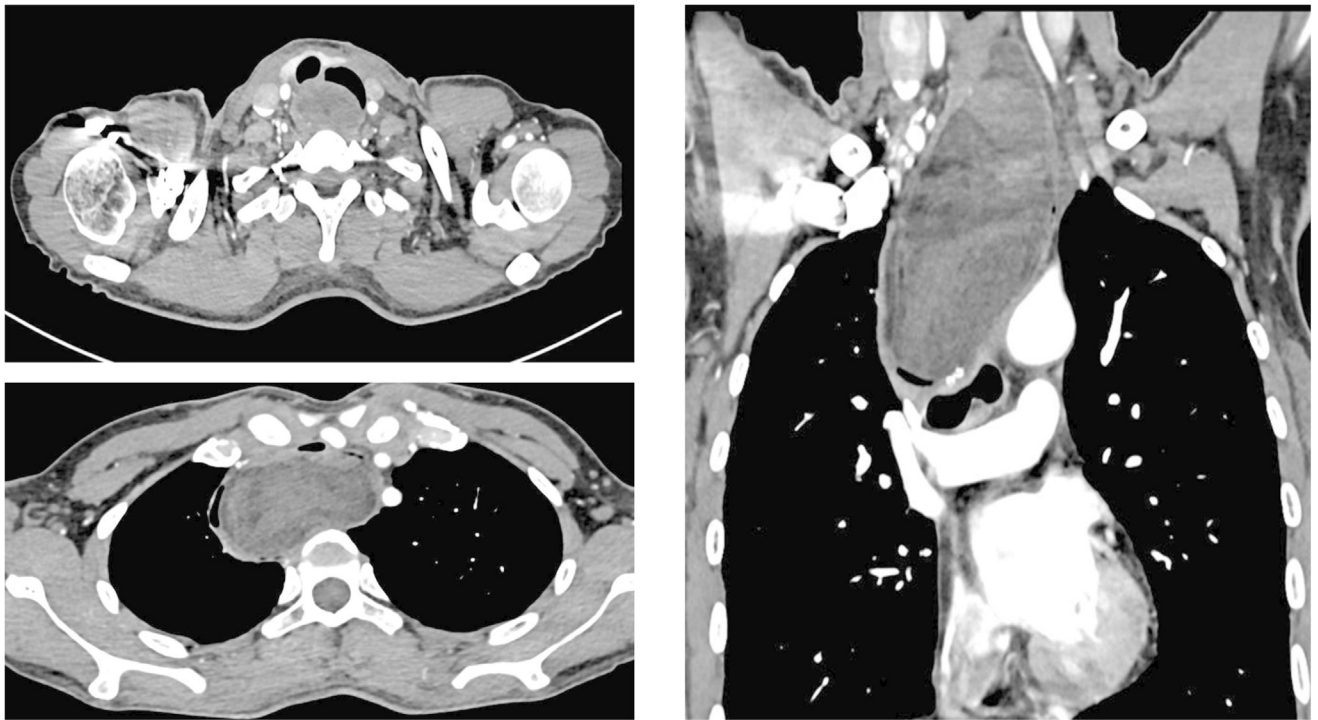


FIGURE 1. Computed tomography scan of the recurrent esophageal liposarcoma.

unresolved.² Given these circumstances, surgical resection emerges as the most suitable course of action. Although endoscopic submucosal dissection has demonstrated success in addressing esophageal nonepithelial tumors, including liposarcomas,³ this approach is deemed unsuitable due to factors such as huge tumor, thoracic adhesion resulting from the initial surgery, and the indistinct demarcation between the tumor and the superior vena cava. In the realm of tumor removal, the efficacy of a cervical incision approach has been well-established, even in cases of recurrence.⁴ However, findings from Ortega and colleagues⁵ raise concerns, as their study revealed that enucleation of liposarcomas located in the posterior mediastinum resulted in positive surgical margins in a noteworthy 63.6% of cases (7 of 11). Taking into account the favorable prognosis associated with the well-differentiated recurrent tumor and the feasibility of achieving en-bloc dissection,^{E1} our multidisciplinary team opted for an esophagectomy, employing a 3-incision technique. This approach was chosen to mitigate the risk of tumor recurrence to the maximum possible extent. Subsequent to the surgery, the patient experienced a favorable recovery, demonstrating the success of the chosen strategy.

CONCLUSIONS

The achievement of successful surgical intervention in this uncommon case of recurrent esophageal liposarcoma underscores a key insight: When dealing with primary esophageal liposarcoma, it is viable to consider tumor enucleation through endoscopic, thoracotomy, or cervical incision approaches, as long as achieving negative surgical margins is possible. However, in the case of tumor recurrence, this outcome underscores that a radical esophagectomy stands as a secure and effective therapeutic measure.

Conflict of Interest Statement

The authors reported no conflicts of interest.

The *Journal* policy requires editors and reviewers to disclose conflicts of interest and to decline handling or reviewing manuscripts for which they may have a conflict of interest. The editors and reviewers of this article have no conflicts of interest.

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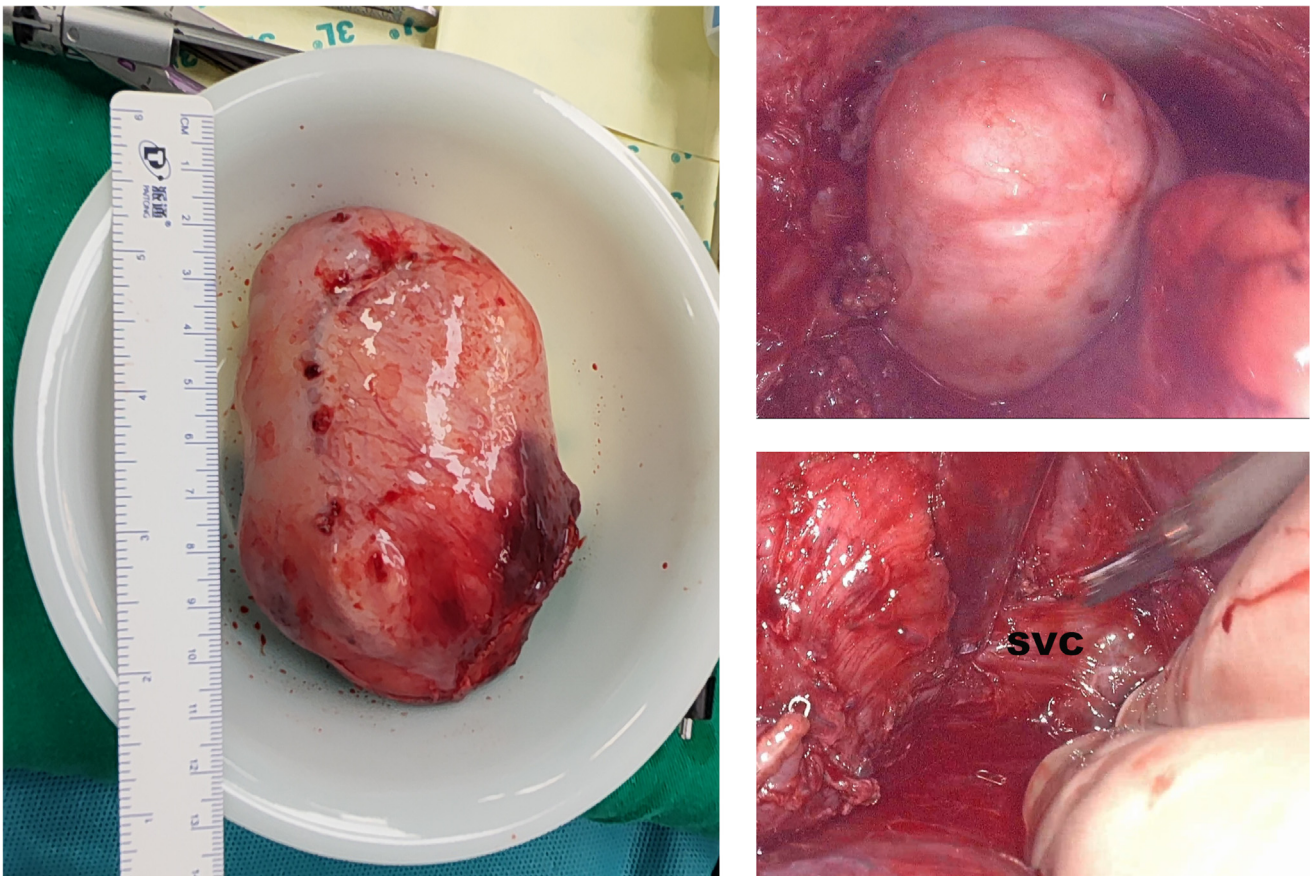


FIGURE 2. Tumor size and images from the surgery.

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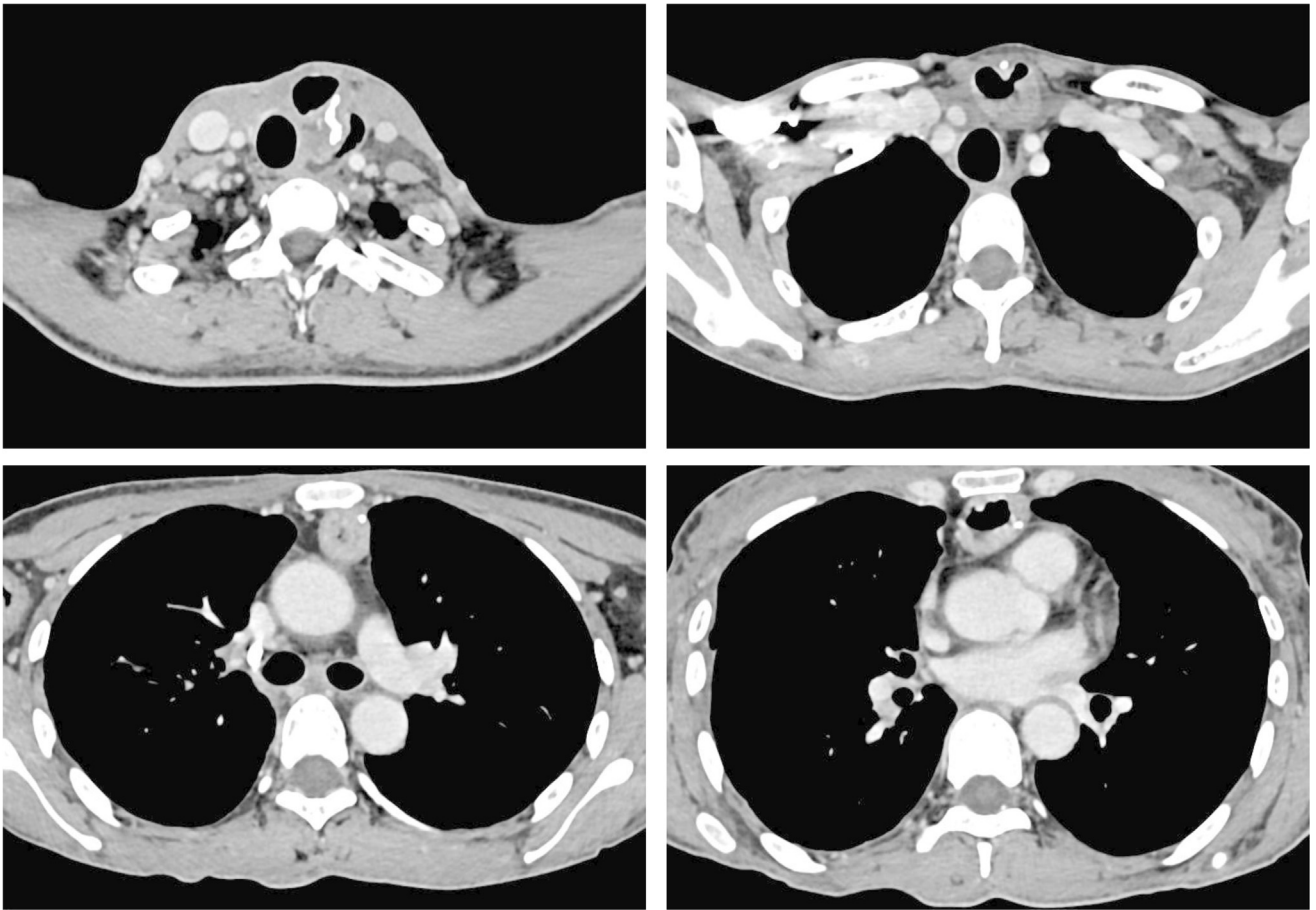


FIGURE E1. Follow-up computed tomography scan of the neck and chest at 1 month after the surgery.