



Thoracoscopic enucleation in the left decubitus position for leiomyoma of the upper thoracic esophagus: Utility of preoperative diagnosis applying endoscopic ultrasound-guided fine needle aspiration

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ABSTRACT

INTRODUCTION: We report a relatively rare case of esophageal leiomyoma in the upper thoracic esophagus enucleated by thoracoscopic procedures. The usefulness of preoperative diagnosis and an adequate surgical approach are described along with a review of the relevant literature.

PRESENTATION OF CASE: A submucosal tumor 45 mm in diameter was detected in the upper thoracic esophagus of a 69-year-old man. The tumor was preoperatively diagnosed from histopathological biopsy under endoscopic ultrasound-guided fine needle aspiration. Thoracoscopic enucleation was therefore preoperatively scheduled under the left decubitus position in consideration of the low risk of malignant disease. Histopathological diagnosis of the resected specimen was benign leiomyoma and patient outcomes were good.

DISCUSSION: The need for preoperative biopsy of esophageal submucosal tumor is a controversial issue. However, preoperative biopsy provided the benefits to decide the operative procedure or confirm adequate resection, and our experience suggested that preoperative biopsy did not adversely influence subsequent enucleation.

CONCLUSION: Precise preoperative diagnosis is necessary to avoid excessive surgery when managing esophageal submucosal tumor.

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This case report has been written in line with the SCARE criteria [1].

1. Introduction

Benign esophageal tumors are relatively rare and reportedly constitute less than 10% of all esophageal neoplasms [2]. Esophageal leiomyoma is the most common benign neoplasm of the esophagus, accounting for about two-thirds of all benign

esophageal tumors and usually occurring in patients between 20 and 60 years old, with a male-to-female ratio of 2:1 [3]. The prevalence of finding this disease has increased with the spread of health examinations and improvements in preoperative imaging diagnosis.

Surgical enucleation of benign esophageal tumors seems to become the consensus position [4,5], and a thoracoscopic approach or per-oral endoscopic tumor resection reduces the invasiveness for patients. However, precise preoperative diagnosis is important because enucleation of gastrointestinal stromal tumor (GIST) with malignant potential resembling leiomyoma is not recommended, given the risk of tumor recurrence. In fact, Nishimura et al. [6] reported recurrence occurring in 4 of 16 patients (25%) who underwent enucleation for GIST of the esophagus. In addition,

Abbreviations: GIST, gastrointestinal stromal tumor; CT, computed tomography; PET-CT, positron emission tomography-computed tomography; EUS-FNA, endoscopic ultrasound-guided fine needle aspiration; ICS, intercostal space.

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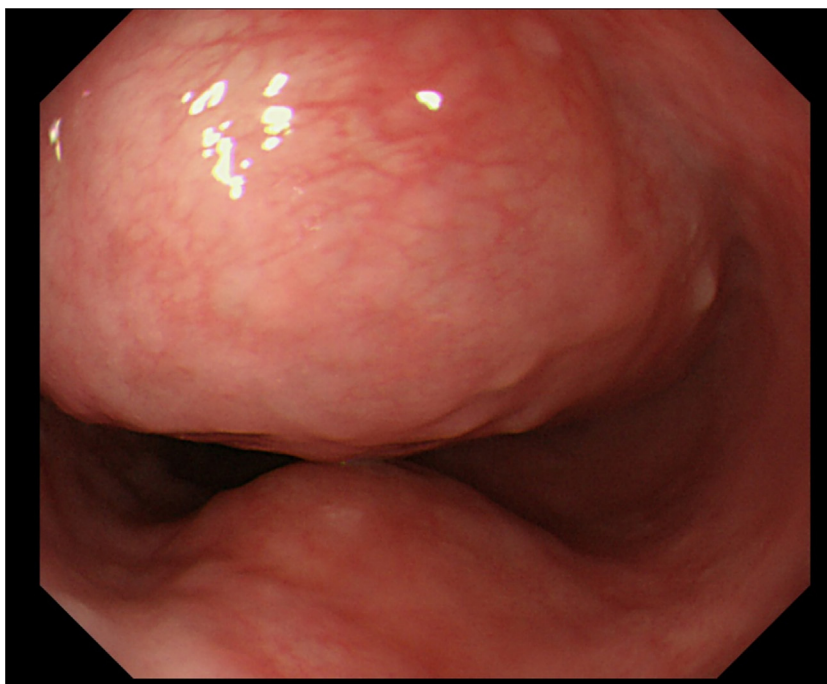


Fig. 1. Endoscopic examination findings.

Endoscopic examination shows submucosal tumor in the upper thoracic esophagus.

no consensus seems to have been reached regarding the optimum patient position in surgery, as the left decubitus or prone position.

In the present report, a case of benign leiomyoma in the upper thoracic esophagus that could be successfully resected by thoracoscopic minimally invasive surgery and the usefulness of preoperative diagnosis and an adequate surgical approach are described along with a review of the relevant literature.

2. Presentation of case

A 69-year-old asymptomatic man with a medical history of chronic atrial fibrillation was referred to our institute after an esophageal submucosal tumor was detected on a periodical upper gastrointestinal endoscopy. Endoscopic examination showed an esophageal submucosal tumor 4 cm in diameter in the upper thoracic esophagus (Fig. 1). In an enhanced upper gastrointestinal series, the lesion appeared as a smooth, rounded filling defect with sharp demarcation resembling a submucosal mass lesion, displacing the adjacent trachea (Fig. 2). Chest computed tomography (CT) revealed a 4.3 × 3.0-cm, round mass lesion arising along the anterior wall of the esophagus in the upper thorax at the level of the aortic arch, appearing as a hypovascular mass without paraesophageal lymphadenopathy (Fig. 3). Positron emission tomography (PET)-CT showed no significant ¹⁸F-fluorodeoxyglucose accumulation in the tumor. Routine laboratory testing showed no abnormalities and no increased levels of tumor markers such as carcinoembryonic antigen or carbohydrate antigen 19-9, and clinical findings were almost normal.

Resection of the tumor was considered necessary because of the gradual growth. To obtain a precise histological diagnosis, endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) was performed. The submucosal tumor was diagnosed histopathologically as benign leiomyoma of the esophagus and thoracoscopic enucleation was therefore scheduled.

Under general anesthesia utilizing a double-lumen tube in the left lateral decubitus position, right thoracoscopy was performed after right lung isolation without artificial pneumothorax. Six trocars were placed, and the camera port was placed at the 8th

intercostal space (ICS) in the middle axillary line. Three 5-mm trocars were placed at the 6th ICS and 8th ICS in the posterior axillary line, and at the 5th ICS in the midaxillary line for the operator. Two 10-mm trocars were placed at the 3rd ICS and 6th ICS in the anterior axillary line for assistance. The tumor was identified on the head side of the azygos arch through the mediastinal pleura. The mediastinal pleura, adventitia of the esophagus and muscularis propria layer were cut longitudinally using an electro-surgical knife to expose the tumor capsule (Fig. 4a). The tumor seemed to mainly arise from the lamina muscularis mucosae, and a round, elastic-hard tumor was successfully enucleated by almost blunt dissection without injury to the mucosa (Fig. 4b). After tumor excision, the mediastinal pleura, adventitia of the esophagus and muscularis propria of the esophagus were closed with interrupted sutures and a 19-Fr chest drain was placed.

Postoperatively, a nasogastric tube was removed on postoperative day 1, and the chest tube was removed on postoperative day 2. Contrast swallow of the esophagus on postoperative day 5 revealed intact smooth esophageal mucosa without leakage or stenosis. The patient started per-oral intake from postoperative day 5. The course after surgery was unremarkable.

Histopathological examination of the resected specimen showed spindle-shaped tumor cells (Fig. 5a), appearing diffusely positive for desmin, but negative for DOG-1, c-KIT, CD34, and S100 on immunohistochemical examination (Fig. 5b). MIB-1 index was approximately 4%. The final pathological diagnosis was leiomyoma of the esophagus with no evidence of malignancy.

3. Discussion

Esophageal leiomyoma arises from the distal third of esophagus in 60% of cases, the middle third in 30%, and the upper third in 10% [7]. This distribution reportedly reflects the distribution of smooth muscle cells in the esophagus [7]. In addition, esophageal leiomyoma is reported to originate from the circular muscle in 74%, lamina muscularis mucosae in 18%, and longitudinal muscles in 8% [8]. Shin et al. [9] described the clinical symptoms of esophageal leiomyoma as dysphagia (12%), epigastric discomfort (8%), dyspep-



Fig. 2. Enhanced upper gastrointestinal series findings.

Enhanced upper gastrointestinal series shows the lesion appearing as a smooth, rounded, filling defect with sharp demarcation as well, as on endoscopy.

sia (6%), chest discomfort (2%), and regurgitation (1%). About half of patients (58%) are asymptomatic, as in the present case.

Surgical treatment of esophageal leiomyoma is considered warranted for symptomatic cases and asymptomatic cases showing increasing size or findings suggestive of malignancy [10]. However, indications for surgical treatment have not been established. Thoracoscopic enucleation is commonly planned for esophageal leiomyoma, whereas thorotomy or esophagectomy are considered for larger tumors. In this case, we performed thoracoscopic enucleation with the aim of achieving minimally invasive surgery. Per-oral endoscopic tumor resection has recently been indicated for small intramural or intraluminal lesions [11], but did not seem appropriate in the present case because of the larger size (diameter, 4 cm).

The need for preoperative biopsy of esophageal submucosal tumor is a controversial issue [10]. Mucosal damage while enucleating the tumor and the potential risks of bleeding, perforation and infection are considered as disadvantages for preoperative biopsy [12,13]. Conversely, EUS-FNA allows leiomyoma to be distinguished from other esophageal submucosal tumor preoperatively. Concerning the indication of EUS-FNA, Vander Noot et al. [14] reported the diagnostic accuracy of EUS-FNA in diagnosing gastrointestinal tract neoplastic lesions as 89%, and Baysal et al. [15] suggested considering EUS-FNA for tumors >3 cm in diameter, possible GIST, or high risk of malignancy. In cases in which a diagnosis of GIST or malignant tumor is possible, preoperative biopsy should be conducted, because recurrence was seen in 4 of 16 patients (25%) who underwent enucleation of GIST of the esophagus [6], while esophagectomy should be considered for GIST regardless of the high surgical stress. Our experience suggests that preoperative biopsy does not adversely influence subsequent enucleation, and preoperative EUS-FNA appears necessary to exclude GIST or confirm adequate resection.

The authors utilize 6 ports for thoracoscopic surgery for esophageal cancer. In the same way, the authors utilized 6 ports for this procedure. On the approach by the left lateral decubitus position, the assistant surgeon must keep the surgical view by compression of right lung. Therefore, the authors adapted 6 ports approach. In this case, we didn't require circumferential mobilization of the esophagus and transection of the azygous vein. It might be possible to reduce one or two ports.

Concerning the body position during thoracoscopic enucleation, the lateral decubitus and prone positions have been introduced in the literature; however, the superiority of these positions may be controversial, as described by Maki et al. [16]. We prefer to select the left lateral decubitus position for lesions in the upper esophagus, and the prone position for lesions in the lower esophagus. As the left lateral decubitus position is good for lymph node dissection of the upper mediastinum for esophageal cancers, a good field of view could be secured over a deep surgical site for the lesion in the upper esophagus. In particular, this position is good for large tumors that cause tracheal deviation, as in the present case. But this position could be adapted to suit the needs of the surgeon, while maintaining the surgical view required for the tumor location.

4. Conclusion

Preoperative EUS-FNA is a useful diagnostic modality for differentiating between benign and malignant submucosal tumor of the esophagus. Precise diagnosis by EUS-FNA can provide informative results for deciding the operative procedure, and thoracoscopic, minimally invasive tumor enucleation can be safely achieved. In addition, the left lateral decubitus position is a good option for upper esophageal tumor enucleation in thoracoscopic surgery.



Fig. 3. CT findings. Contrast-enhanced chest computed tomography reveals a 4.3 × 3.0-cm mass (arrow) displacing the adjacent trachea, arising along the anterior wall of the esophagus in the upper thoracic part at the level of the aortic arch.

Conflicts of interest

Mitsutoshi Ishii and other co-authors have no conflict of interest.

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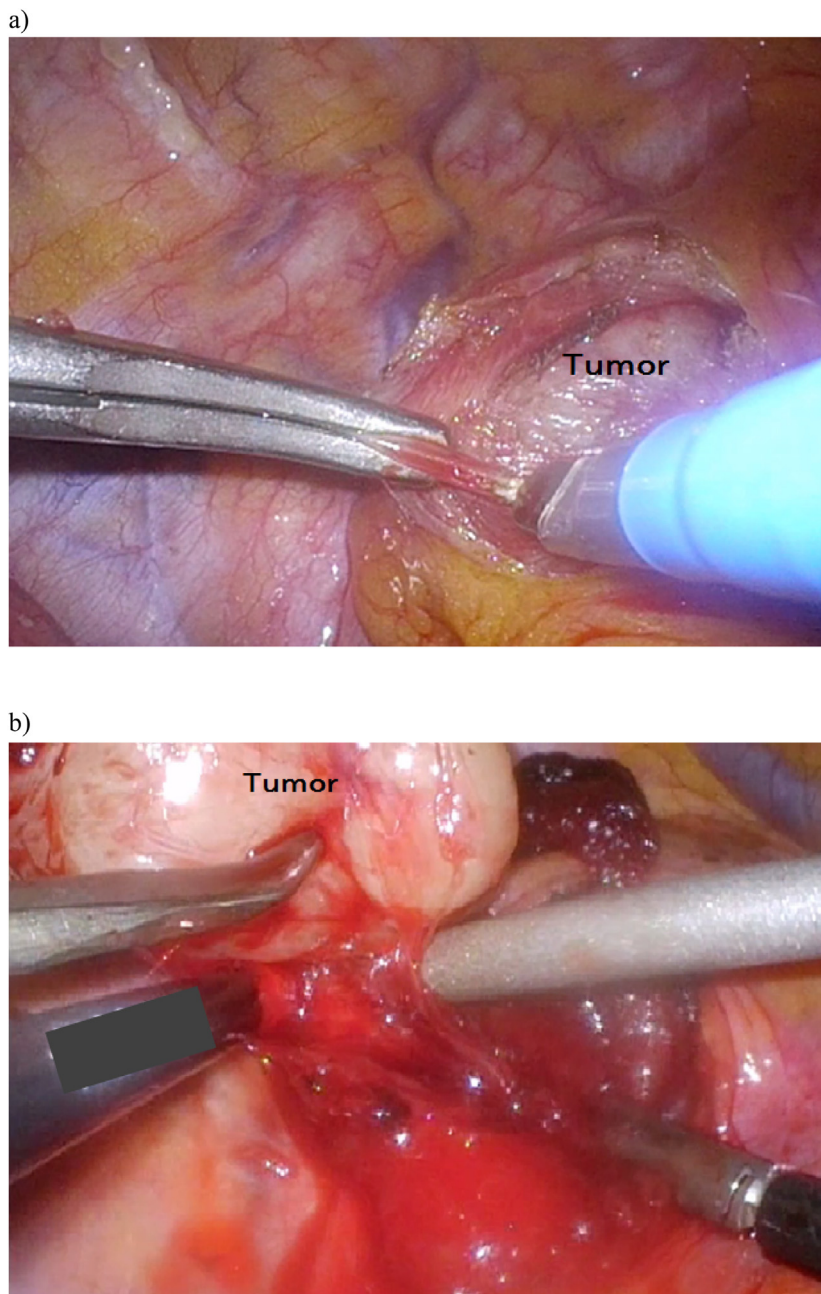


Fig. 4. Intraoperative findings.

a) The capsule of the tumor is exposed after cutting the mediastinal pleura, and the adventitia of the esophagus and muscularis propria layer are cut longitudinally using electrocautery.

b) The tumor has been completely enucleated by almost blunt dissection without injuring the esophageal mucosa.

Ethical approval

This is not a research study.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Authors contribution

Mitsutoshi Ishii wrote the manuscript. Shinsuke Takeno, Takahiro Nishida and Atsushi Nanashima reviewed critically the manuscript. Yoshimasa Kubota and Hiroshi Kawakami conducted

EUS-FNA. Yoshiko Umekita and Yutaka Akiyama conducted pathological diagnosis.

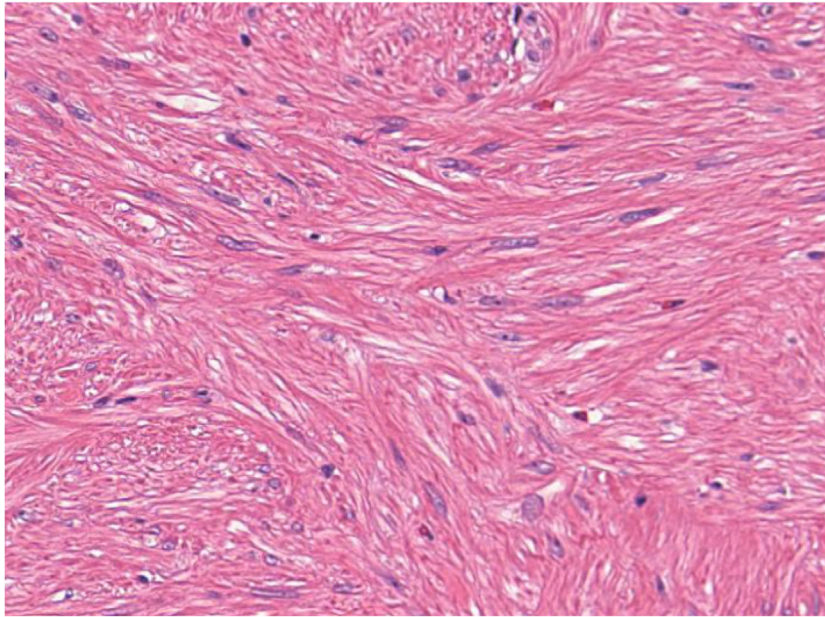
Registration of research studies

This is not a research study.

Guarantor

This is not a research study. All authors read and approved the final manuscript. Therefore, all authors are responsible for this article.

a)



b)

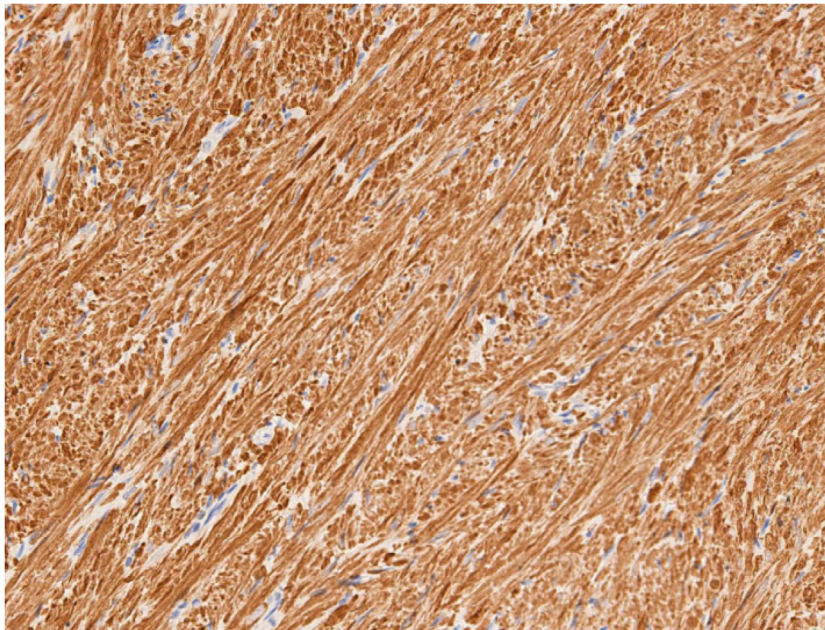


Fig. 5. Microscopic findings.

- a) Hematoxylin-eosin staining shows proliferation of spindle-shaped tumor cells forming interlacing and fascicles. Cellularity is relative low, and neither coagulation necrosis nor high mitotic activity of the tumor were noted.
- b) Immunohistochemically, spindle-shaped tumor cells were diffusely positive on desmin staining.

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