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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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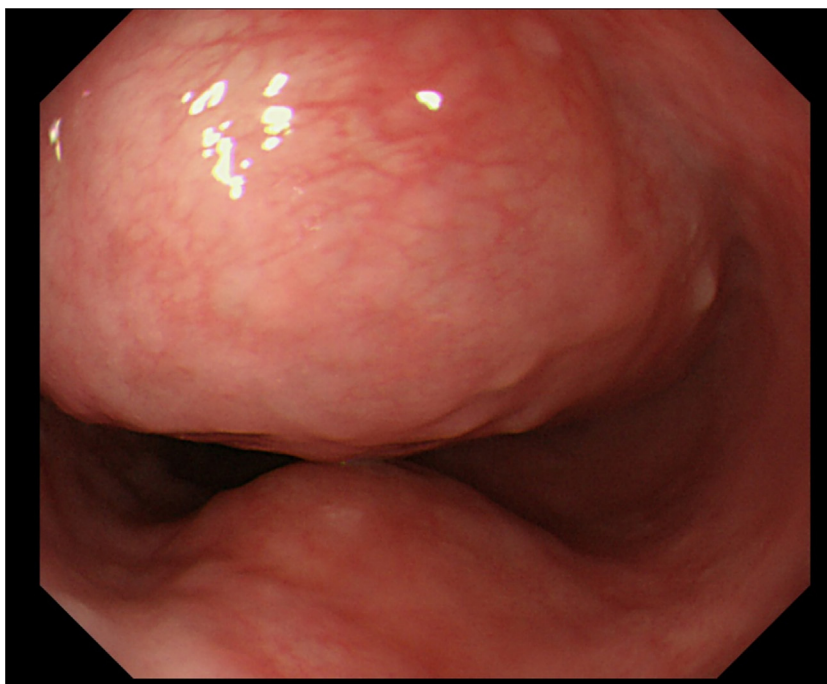


Fig. 1. Endoscopic examination findings.

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-

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