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CASE REPORT

Successful thoracoscopic resection of an esophageal bronchogenic cyst

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Abstract

Esophageal bronchogenic cysts are rare, and since a limited number of cases has been reported, the treatment plan for asymptomatic patients is difficult. A 55-year-old man without any symptoms visited our hospital for further examination of an esophageal mass detected on simple computed tomography. Upper endoscopy showed the protruding, submucosal mass covered by normal mucosa, and endoscopic sonography confirmed hypoechoic lesions originating from the muscularis propria. The patient was diagnosed as having an esophageal duplication cyst and underwent thoracoscopic resection. Pathological findings were consistent with an esophageal bronchogenic cyst. The patient was discharged without any problems on the 6th postoperative day. Upper endoscopy was performed 6 months after surgery, and no evidence of esophageal diverticula or narrowing was present. In conclusion, early thoracoscopic resection of esophageal bronchogenic cysts, before appearance of the symptom, can be a considerable treatment option because it is less invasive and may be advantageous for obtaining a definitive diagnosis in patients who are candidates for safe surgical resection.

Keywords Esophageal bronchogenic cyst · Esophagus duplication cyst · Thoracoscopic resection

Introduction

Esophageal bronchogenic cysts (EBCs) and esophageal duplication cysts (EDCs) are both benign bronchopulmonary malformations that arise from abnormal budding of the primitive tracheobronchial tube [1]. They are clinically very similar, making accurate diagnosis difficult without surgical resection [2]. The treatment plan for patients without symptoms such as dysphagia and chest pain, is unclear. However, owing to the development of a thoracoscopic procedure [3], thoracoscopic resection of esophageal benign tumors is feasible and safe. Herein, we present a case of an esophageal bronchogenic cyst successfully removed by the thoracoscopic procedure.

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Case

A 55-year-old man presented to our hospital for further examination of an esophageal mass detected on a simple computed tomography (CT) scan. He did not complain of any symptoms such as dysphagia and chest pain. Physical examination and laboratory findings were unremarkable. The chest radiograph did not show any tumors. Enhanced CT showed a well-defined homogenous lesion with 30 mm in size along the right side of the middle esophagus (CT number: 10–51 HU) (Fig. 1a, b). Magnetic resonance imaging also showed a 30-mm lesion with cystic fluid, but without any solid tumors, adhered to the esophagus (Fig. 1c, d).

Upper endoscopy showed that the protruding, submucosal mass was covered by normal mucosa at 28 cm from the incisor tooth (Fig. 2a, b). Endoscopic sonography confirmed hypoechoic lesions originating from the muscularis propria, indicating that the lesions were located intramuscularly in the esophagus (Fig. 2c). We did not perform a fine-needle aspiration biopsy and endobronchial ultrasound-guided trans-bronchial needle aspiration (EBUS-TBNA) by trans-bronchial puncture because it could cause infection of the cysts or bleeding [4].

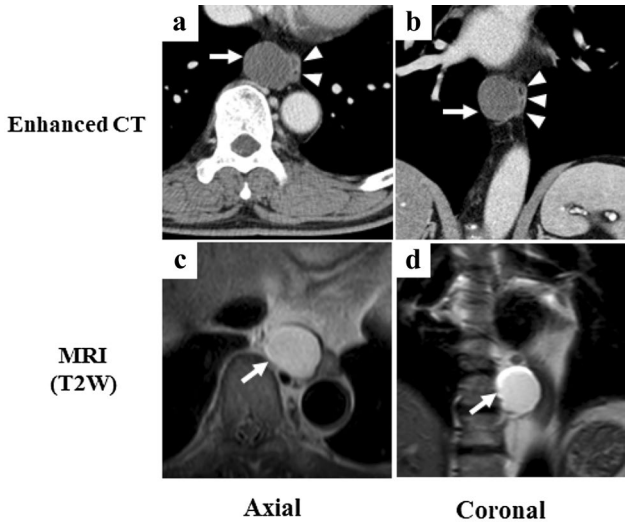

Fig. 1 **a**, **b** Enhanced CT showed a well-defined homogenous lesion with 30 mm in size along the right side of the middle esophagus (arrows). The esophagus was compressed by the cyst (arrowheads). **c**, **d** T2-weighted magnetic resonance imaging also showed a 30-mm lesion with cystic fluid, but without any solid tumors, adhered to the esophagus (arrows)

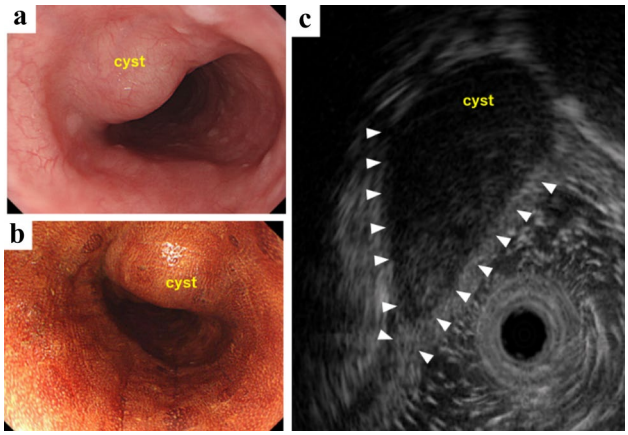

Fig. 2 **a**, **b** Upper endoscopy showed that the protruding, submucosal mass covered by normal mucosa in the anterior of lower esophagus (28 cm from the incisor tooth) (**a** conventional white light, **b** Lugol's iodine stain). **c** Endoscopic sonography confirmed hypoechoic lesions (arrow) originating from the muscularis propria (arrowheads), indicating that the lesions were located intramuscularly in the esophagus

We first suspected EDC because the cysts laid within the esophageal wall and were covered by two muscular layers. The patient was informed about several treatment options including surgical resection and conservative follow-up and decided to undergo thoracoscopic resection. The patient was intubated with a single lumen endotracheal tube and moved in the prone position. Four thoracoscopic ports were introduced. The AirSeal® access port (ConMed, Utica, NY) and the camera port, ENDOPATH XCEL Bladeless 10 mm Trocar (Ethicon, Johnson & Johnson, Japan), were placed at the seventh and 10th intercostal spaces, posterior to the tip of scapula, respectively. Two additional XCEL 5-mm Trocars were placed at the fifth and seventh intercostal spaces in the posterior axillary line and were used for the majority of the procedure (Fig. 3a). A 5-mm HD flexible videoscope (VISERA ELITE platform, Olympus) was used, and the cyst was observed protruding in the middle mediastinum, which was capsulated within the mediastinal layer. The membrane, esophageal adventitia and the part of esophagus muscularis propia, around the cyst was cut, and the cyst was completely resected without breaking the cyst using downward retraction to the caudal side. An endoscopic observation during the surgery was also effective to perform the safe surgical resection of the tumor, especially protruding to the both sides (inside and outside) of esophagus. The right vagus nerve was observed running beside the cyst and carefully conserved during resection. As the muscle layer was partially injured after the resection, we closed the mediastinal membrane with the muscle layer together by 3-0vicryl (absorbable, braided suture, Ethicon) (Fig. 3b–d). The cyst was extracted in the plastic bag safely through the 3 cm thoracotomy of 12 mm port.

The operation time was 134 min, and the estimated blood loss was minimal. The cyst was solid but elastic and contained a milky white material with zelatin (Fig. 4a). Pathological findings indicated the presence of cartilage or respiratory glands in the ciliated columnar epithelium, and the patient was diagnosed as having EBC (Fig. 4b).

The patient started eating on the second postoperative day and was discharged with uneventful recovery on the sixth postoperative day. The patient was able to consume a regular diet and experienced no symptoms related to esophageal diverticula or narrowing. No evidence of esophageal diverticula or narrowing was present on upper endoscopy performed 18 months after surgery.

Discussion

Bronchogenic cysts form as the result of abnormal budding of the primitive tracheobronchial tree, typically occurring in the mediastinum [2, 5], and most involve the lung parenchyma, while periesophageal tumors are very rare [6]. EBCs and EDCs are both benign bronchopulmonary malformations and differential diagnosis between them without pathological analysis of surgically resected tumors is difficult. Ko et al. reported seven cases of EBC, none of which were diagnosed before surgery [7]. EBUS-TBNA by transbronchial puncture has been reported as a less invasive diagnostic tool of suspected intrathoracic tumor [8]. On the other hand, there was a report of pneumoniae following by EBUS-TBNA

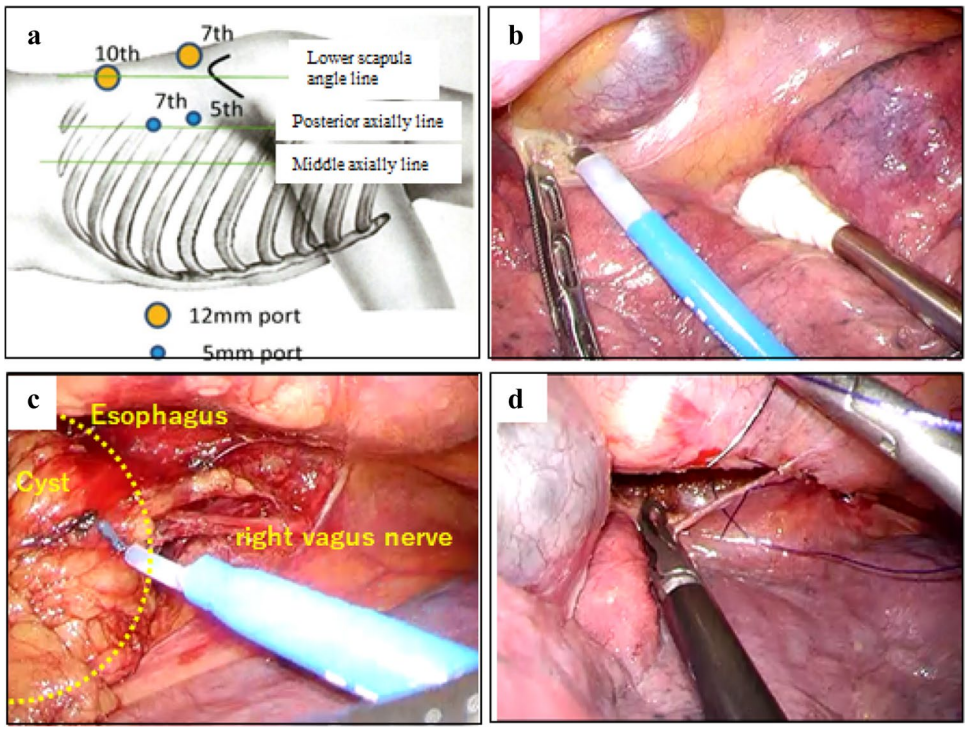

Fig. 3 **a** The placement of four ports. **b** The cyst was observed protruding in the middle mediastinum, which was capsulated within the mediastinal layer. **c** After the cyst resection. The left vagus nerve was observed running beside the cyst. **d** Closure the mediastinal membrane with the muscle layer

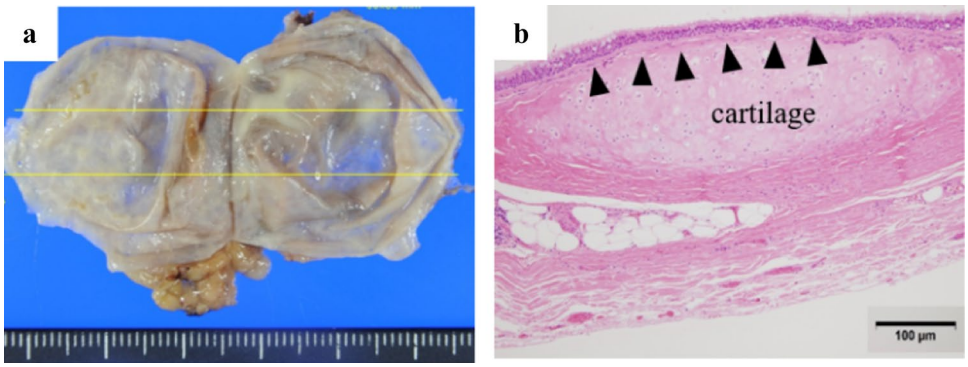

Fig. 4 **a** Macroscopic findings of the resected cyst. **b** Pathological findings of the cyst indicated the presence of cartilage or respiratory glands in the ciliated columnar epithelium (arrowheads) (HE × 100)

for bronchogenic cyst [3]. We discussed deeply with the patient about the risk and benefits of diagnostic fine needle biopsies, however, he chose the complete surgical resection without the preoperative biopsies because they might have not been sufficient for definite diagnosis regardless of complication risks. Fortunately, the treatment plans for these esophageal benign tumors are similar. If patients present with any symptoms, such as dysphagia or chest pain, early surgical resection should be considered. However, the optimal treatment strategy for small, asymptomatic tumors remains unclear, with both conservative follow-up and surgical resection as viable options.

Bronchogenic cysts, especially EBCs, ultimately become symptomatic over time, and symptoms can sometimes be serious because of the compression of adjacent structures [5, 9, 10]. Malignancy within these cysts is rare, but has been reported [11]. The surgical procedure is also more complicated and hazardous when the cyst is symptomatic because of infection or effects on adjacent organs [5]. St-Georges et al. reported that major operative complications occurred in 40.7% of patients with bronchogenic cysts who underwent posterolateral thoracotomy, all of whom were symptomatic preoperatively. Thoracoscopic surgery is less invasive and has recently become a safe procedure because of advances in magnified viewing and surgical devices. Several reports showed the advantages of thoracoscopic resection for definitive diagnosis and prevention of serious complications [7]. Although open procedures are truly superior in the tactile sensation of the tissue to thoracoscopic procedures, precise surgical manipulation with the deep understanding of tumor location and the surrounding tissues with the HD videoscope can be expected to overcome the disadvantage. In this case

we needed the skilled technique, because the tumor was originating from the right side of esophagus, where located at the far side from the protruding vertebra (Fig. 3b, c). With the help of good view of the 5-mm HD flexible videoscope and fined surgical device such as hook type electric knife enabled us to perform the safe manipulation and complete tumor resection even in such a complicated case. Moreover, the mobilization of the esophagus with taping technique was indispensable for excellent surgical view and maintaining for the good surgical orientation. The intra-operative esophagoscopy with balloon dilator assistance is also useful to the exact localization of the tumor and the navigation for surgery. We believed that the thoracoscopic procedure was also useful in the minimum injury of muscle layer in this case. Because here have been reports of cases with recurrence following an incomplete resection [12], a complete resection of the cyst is essential to prevent recurrence. Based on these reasons, we considered that the complete surgical resection, especially before the appearance of symptom, is pivotal for undiagnosed esophageal cyst, regardless of the size of tumor. Moreover, the thoracoscopic surgery should be considered after careful and comprehensive evaluation of the tumor, and also after constructing the feasible surgical strategy. In our case, we adopted the same method of thoracoscopic surgery of esophageal cancer including the prone position of the patient and appropriate position of trocars for a good visual field. In addition, hook type electric scalpel enabled the fine dissection of the membrane around the cyst and along the vagus nerve, which were the pitfall for complete resection of the cyst. We believed that these strategies were helpful for the cases which are suspicious of malignancy or giant cyst. Ota et al. reported the asymptomatic case of bronchogenic cyst originating from diaphragm successfully removed via thoracoscopic surgery and suggested the surgical resection of bronchogenic cyst was preferred to conservative follow-up in definitive diagnostic, risk of surgery after appearance of symptoms and malignancy potential [13]. Similar results were observed in our case, indicating that thoracoscopic surgery was safe and effective in asymptomatic patients.

Conclusion

As the preferred treatment for undiagnosed, asymptomatic esophageal cysts, early thoracoscopic resection of the cyst, especially before appearance of symptom, can be a considerable treatment option because it is less invasive and advantageous for definitive diagnosis and safe surgical resection.

Declarations

Conflict of interest All authors declare that they have no competing interests.

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

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