Patient Report

# Thoracic Esophagus Cancer Revealing a Tracheal Diverticulum

Takuki Yagyu,\* Hiroaki Saito,† Yusuke Kono,† Yuki Murakami,† Hirohiko Kuroda,† Tomoyuki Matsunaga,† Yoji Fukumoto,† Shuichi Takano,† Tomohiro Osaki† and Yoshiyuki Fujiwara†

\*Department of Surgery, Tottori Prefectural Central Hospital, Tottori 680-0901, Japan and †Division of Surgical Oncology, Department of Surgery, School of Medicine, Tottori University Faculty of Medicine, Yonago 683-8503, Japan†

#### **ABSTRACT**

Tracheal diverticulum is rarely encountered in a clinical setting since almost all patients are asymptomatic. However, its presence may become a problem during esophageal cancer operations in terms of anesthesia and lymph node dissection of superior mediastinum lymphadenectomy. A 70-year-old man with esophageal cancer was referred to our hospital. During thoracoscopic subtotal esophagectomy, we found a cystic lesion connected to the right posterior wall of the trachea. We evaluated the preoperative computed tomography scan during surgery and made a diagnosis of tracheal diverticulum because of the presence of paratracheal air cysts, which had not been noticed preoperatively. It was resected by a linear stapler and the postoperative course of the patient was uneventful. A careful preoperative evaluation of computed tomography and operation are necessary to avoid injury of tracheal diverticulum during thoracoscopic esophagectomy for esophageal cancer revealing a tracheal diverticulum.

**Key words** esophageal cancer; thoracoscopic esophagectomy; tracheal diverticulum

Tracheal diverticulum is a benign entity characterized by single or multiple invaginations of the tracheal wall. Although it is rarely encountered in a clinical setting since almost all patients are asymptomatic, the prevalence of tracheal diverticulum has been estimated to be about 1% according to autopsies. 1-3 Tracheal diverticulum is typically located at the right posterior wall of the trachea a few centimeters above the tracheal bifurcation. This region is critical for the surgery of esophageal cancer because there are right recurrent nerve lymph nodes (#106 recR), which should be dissected at this region. Injury to tracheal diverticulum during the operation for esophageal cancer might put patients in serious condition after operation. Therefore, careful evaluation of preoperative

computed tomography (CT) and operation are necessary to avoid injury of tracheal diverticulum.

In this case report, we present an esophageal cancer patients with tracheal diverticulum in whom thoracoscopic esophagectomy combined with resection of tracheal diverticulum has been performed.

#### PATIENT REPORT

An asymptomatic 70–year–old man with a history of distal gastrectomy for gastric ulcer was diagnosed with esophageal cancer at a neighboring hospital and was referred to our hospital. His other past medical history was appendicitis, diabetes mellitus, and chronic obstructive pulmonary disease (COPD). Under preoperative diagnosis of stage II (T2N0M0) squamous cell carcinoma of thoracic esophagus, thoracoscopic subtotal esophagectomy and open total remnant gastrectomy were performed, while pedunculated jejunum and esophageal stump were anastomosed at the left neck after two course of neoadjuvant chemotherapy (5–fluorouracil 800 mg/m $^2$  × 5, cisplatin 80 mg/m $^2$ ).

During superior mediastinum lymphadenectomy, we found a cystic lesion connected to the right posterior wall of the trachea at the Th2 level (Figs. 1a and b). At that time, we evaluated the preoperative CT scan again and made a diagnosis of tracheal diverticulum because of the presence of paratracheal air cysts, which were not noticed preoperatively (Figs. 2a and b). Although we performed superior mediastinum lymph node dissection without obvious injury of tracheal diverticulum, we resected it using a linear stapler under consideration of possibility of thermal injury by ultrasonically activated scalpel.

Macroscopically, the resected tracheal diverticulum measured  $13 \times 13$  mm (Fig. 3a). Microscopic examination of the resected and H&E-stained specimens revealed that the diverticular wall was covered by pseudostratified ciliated epithelium. It had no cartilage or smooth muscle and was diagnosed as tracheal diverticulum (Figs. 3b and c). The postoperative course of the patient was uneventful, and he was discharged on day 37.

Corresponding author: Hiroaki Saito, MD, PhD sai10@med.tottori-u.ac.jp
Received 2017 June 14
Accepted 2017 July 27
Abbreviations: CT, computed tomography; COPD, chronic obstructive pulmonary disease

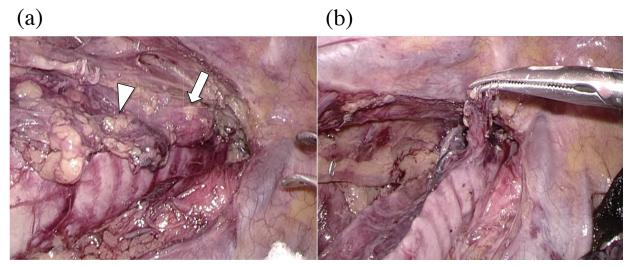
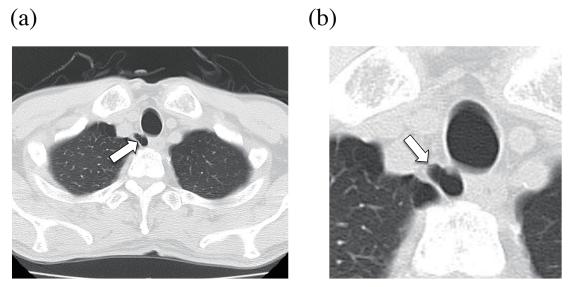


Fig. 1. Intraoperative finding. (a) The tracheal diverticulum during superior mediastinum lymphadenectomy. The tracheal diverticulum (arrow) had a similar appearance to the dissected lymph nodes (arrow head). (b) Tracheal diverticulum connected to the right posterior wall of the trachea.



**Fig. 2.** CT image. (a) Computed tomography image showing a paratracheal air cyst located at right posterior wall of trachea (arrow). (b) The paratracheal air cyst (arrow) seems to have a septal wall between the lungs. These seems to be a communication between trachea and paratracheal air cyst.

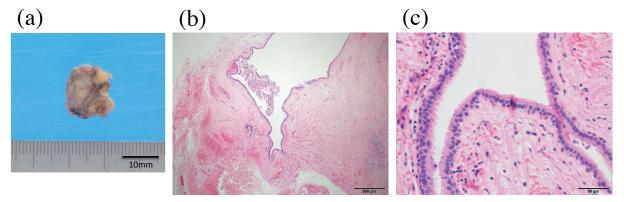


Fig. 3. Macroscopic and microscopic image. (a) Surgical specimen shows a diverticulum measured 13 mm  $\times$  13 mm. (b) There is no cartilage and smooth muscle in the tracheal diverticulum (hematoxylin and eosin stain). Bar = 500  $\mu$ m. (c) The wall of diverticula was covered by pseudostratified ciliated epithelium. Bar = 50  $\mu$ m.

Table 1. Summary of reported cases of tracheal diverticulum with esophageal cancer

Case	Age (years)	Sex	Location	Level	Operation method	Preoperative diagnosis	Resection	Adverse event
110	74	F	Right posterior	Th1	Thoracoscopic	+	-	-
211	63	M	Right posterior	Above the tra- cheal bifurcation	Open	-	-	Difficulty of one- lung ventilation
3 (Our case)	70	M	Right posterior	Th2	Thoracoscopic	-	+	-

F, female; M, male; Th, thoracic vertebrae.

### **DISCUSSION**

Tracheal diverticulum was first reported by Rokitansky in 1838. This diverticulum can be divided into two types: congenital and acquired. The congenital type is considered to be a malformed supernumerary lung and generally consists of respiratory cartilage, smooth muscle and epithelium. It is usually located 4-5 cm below the vocal cords. On the other hand, the acquired type can be further divided into two types: pulsion and traction. The pulsion type is caused by increased intraluminal pressure by repeated coughing or obstructive pulmonary disease combined with vulnerability of the bronchial wall, while the traction type is caused by inflammation in the thoracic cavity. Unlike the congenital type, acquired type tracheal diverticulum is often composed of respiratory epithelium without any cartilaginous or smooth muscular element. Although the predilection sites of both congenital and acquired type tracheal diverticula is the right posterolateral wall of the trachea, the pulsion type tends to be located higher than the congenital type.<sup>4-6</sup> Because of his past history of COPD and lack of cartilage and smooth muscle, our case is considered to be pulsion type tracheal diverticulum.

In our case, patient suffered from esophageal cancer and underwent thoracoscopic subtotal esophagectomy for the treatment of esophageal cancer. There are two points to be considered for a patient with tracheal diverticulum during thoracic esophageal cancer operation. First, it is likely that tracheal diverticulum might be affected by some trouble with anesthesia during operation. In fact, ventilatory insufficiency and injury of the broncus, which result in pneumomediastinum, have been reported thus far.<sup>7-9</sup> Second, tracheal diverticulum sometimes induces respiratory tract infection, which results in adhesion of surrounding tissue. This phenomenon makes lymph node dissection extremely difficult and increases the risk of injury of surrounding organs. Therefore, detailed evaluation of preoperative images is indispensable to perform the operation safely. However, it is difficult to preoperatively diagnose tracheal diverticulum without the knowledge of the tracheal diverticulum because most tracheal diverticulum is asymptomatic. Tracheal diverticulum is usually incidental on radiographic or CT findings. Paratracheal air cysts were visible in only 14% if the study subjects on chest radiography.<sup>5</sup> The optimal modality for diagnosis seems to be a thin-slice CT of the trachea and 3-dimentional reconstruction. In our case, there was a paratracheal air cyst in preoperative CT image. However, we were not able to make a preoperative diagnosis of tracheal diverticulum because of the lack of knowledge of tracheal diverticulum. Because it has been reported that the overall prevalence of tracheal diverticulum has been estimated to be 1% in an autopsy series, it is important to carefully evaluate the presence of tracheal diverticulum in preoperative CT imaging. It is extremely important to protect tracheal diverticulum during esophagectomy and lymph node dissection since tracheal diverticulum is fragile and risky for perforation or even esophago-tracheal fistula. Recently, energy devices such as ultrasonically activated scalpel and vessel sealing system are frequently used in thoracoscopic esophagectomy. In our case, we resected the tracheal diverticulum using a linear stapler considering the possibility of thermal damage to tracheal diverticulum by ultrasonically activated scalpel. There has been two case reports of esophageal cancer revealing a tracheal diverticulum in which thoracic esophagectomy was performed thus far (Table 1).10,11 In all cases, the tracheal diverticulum was located at the right posterior wall of the trachea and the level of upper thoracic spine. Unfortunately, there was only one case in which preoperative diagnosis of tracheal diverticulum was possible. In case 2, the anesthesiologist had difficulty performing one-lung ventilation, because the bronchial blocker was caught in the tracheal diverticulum.

In those patients, resection of tracheal diverticulum was not performed and the postoperative courses of those patients were uneventful. Therefore, it remains unclear as to whether tracheal diverticulum should be removed during esophageal cancer operations or not. Because injury of trachea put patients in serious condition after operation, we believe that tracheal diverticulum should be resected especially in thoracoscopic esophagectomy if there is some possibility of injury. Furthermore, a shortened period of tracheal intubation after operation is recommended to avoid ventilator associated injury or pneumonia.

The authors declare no conflicts of interest.

## **REFERENCES**

- 1 Mackinnon D. Tracheal diverticula. J Pathol Bacteriol. 1953;65:513-7. PMID: 13062051.
- 2 Haghi Z, Towhidi M, Fattahi H, Lari SM. Right paratracheal air cyst (tracheal diverticulum). Respir Care. 2009;54:1409-11. PMID: 19796423.
- 3 Goo JM, Im JG, Ahn JM, Moon WK, Chung JW, Park JH, et al. Right paratracheal air cysts in the thoracic inlet: clinical and radiologic significance. AJR Am J Roentgenol. 1999;173:65-70. PMID: 10397101.
- 4 Ettman IK, Keel DT, Jr. Tracheal diverticulosis. Radiology. 1962;78:187-91. PMID: 13890919.

- 5 Sato T, Sasaki Y, Yamasaki M, Aragaki M, Mae S, Irie T, et al. A right paratracheal air cyst caused by tracheal diverticula. Intern Med. 2010;49:315-9. PMID: 20154437.
- 6 Charest M, Sirois C, Cartier Y, Rousseau J. Infected tracheal diverticulum mimicking an aggressive mediastinal lesion on FDG PET/CT: an interesting case with review of the literature. Br J Radiol. 2012;85:e17-21. PMID: 22190757.
- 7 Frenkiel S, Assimes IK, Rosales JK. Congenital tracheal diverticulum. A case report. Ann Otol Rhinol Laryngol. 1980;89:406-8. PMID: 6776862.
- 8 Dinner M, Ward R, Yun E. Ventilation difficulty secondary to a tracheal diverticulum. Anesthesiology. 1992;77:586-7. PMID: 1519795.
- 9 Moller GM, ten Berge EJ, Stassen CM. Tracheocele: a rare cause of difficult endotracheal intubation and subsequent pneumomediastinum. Eur Respir J. 1994;7:1376-7. PMID: 7925920
- 10 Zhang M, Wang H, Wu WB, Zhang H. Esophageal cancer revealing a tracheal diverticulum. J Thorac Dis. 2015;7:E85-7. PMID: 25973258.
- Miyakita H, Ozawa S, Oguma J, Kazuno A, Yamasaki Y, Iwasaki M. A case of cancer of the thoracic esophagus associated with a tracheal diverticulum. Nihon Rinsho Geka Gakkai Zasshi. 2014;75:3020-23.