

[CASE REPORT]

Aorto-esophageal Fistula Occurring during Lung Cancer Treatment: A Case Treated by Thoracic Endovascular Aortic Repair

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Abstract:

A 63-year-old man had received chemoradiotherapy 7 years ago for stage IIIA pulmonary adenocarcinoma of the left lower lobe and stereotactic irradiation 3 years ago for stage IA pulmonary squamous cell carcinoma of the left upper lobe. An esophageal stent was placed because of esophageal narrowing caused by tumor invasion. Five months later, he was diagnosed with an aorto-esophageal fistula. Because invasive surgery posed challenges, thoracic endovascular aortic repair (TEVAR) was performed. We report this rare case of aorto-esophageal fistula treated using TEVAR. However, the therapeutic effect was temporary. Further studies investigating the indications for TEVAR are warranted.

Key words: TEVAR, aorto-esophageal fistula, lung cancer

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Introduction

Aorto-esophageal fistula is a very rare condition, and its standard treatment has not yet been established. Furthermore, the prognosis is extremely poor (1, 2). The condition arises primarily because of infection, and develops between the esophagus and aorta, leading to massive hemorrhaging and shock. Treatment of aorto-esophageal fistula requires surgery, focusing on removal of the infection source and revascularization, as well as prevention of reinfection. However, the patient's general condition is often poor, and invasive surgery carries a very high risk. Therefore, fistula closure has recently been attempted using thoracic endovascular aortic repair (TEVAR), which is a low-invasive procedure. Nevertheless, the clinical outcomes after TEVAR are not necessarily good. We herein report a rare case of aorto-esophageal fistula treated using TEVAR.

Case Report

A 63-year-old man was admitted urgently to our hospital with hematemesis. The patient had used to previously smoked 37 packs of cigarettes per year. Chemoradiotherapy comprising 2 Gy×30 fr, (total 60 Gy), 2 courses of carboplatin (AUC 2) plus paclitaxel (50 mg/m²) weekly, and 2 courses of carboplatin (AUC 6) plus paclitaxel (200 mg/m²) every 3 weeks had been administered 7 years previously for stage IIIA pulmonary adenocarcinoma of the left lower lobe. Three years ago, the patient had undergone stereotactic irradiation (12.5 Gy×4 fr, total 50 Gy) for stage IA pulmonary squamous cell carcinoma of the left upper lobe (Fig. 1).

The cancer recurred 2 years ago, at which point he was administered two chemotherapy regimens comprising 6 courses of carboplatin (AUC 5) plus tegafur-gimeracil-oteracil (100 mg/day, days 1-14) every 3 weeks and 2

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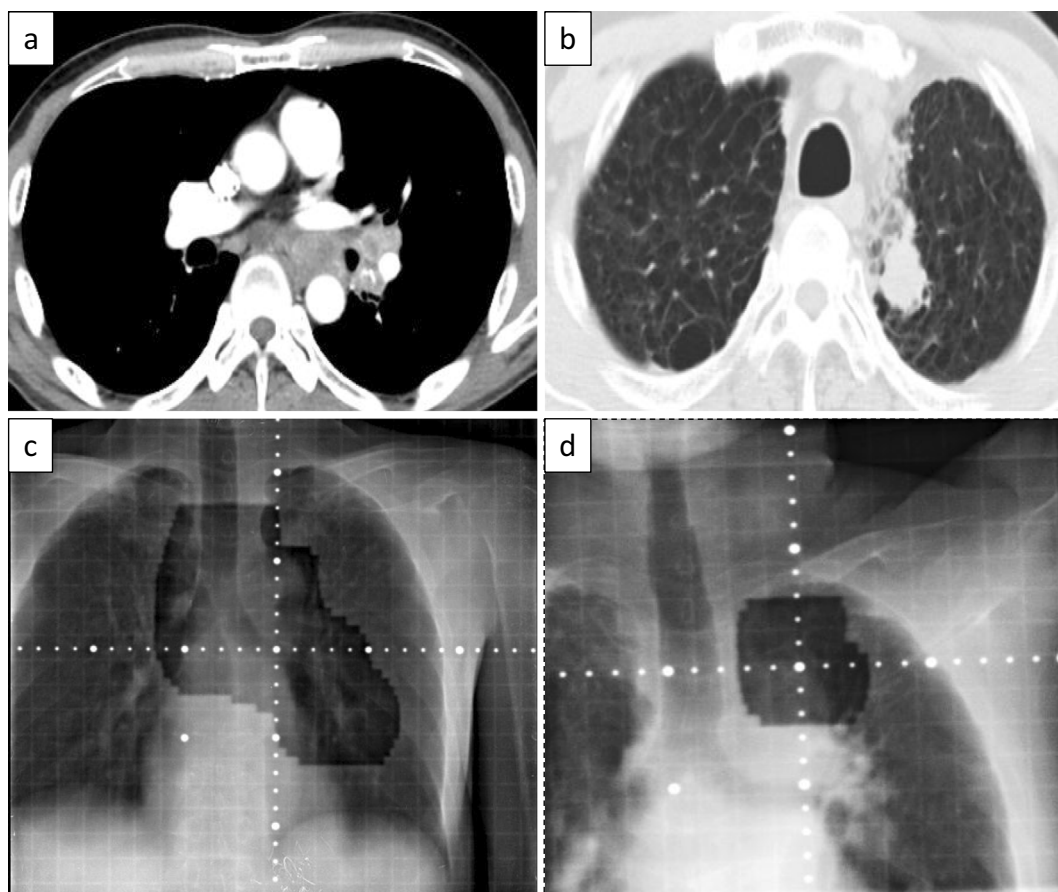


Figure 1. a: Chest computed tomography seven years prior to the admission showed enlarged mediastinal and hilar lymph nodes. b: Chest computed tomography three years prior to the admission showed a tumor in the left upper lobe S¹⁺². c, d: Chest X-ray shows the radiation field of stereotactic irradiation for each treatment.

courses of carboplatin (AUC 4.5) plus nab-paclitaxel (100 mg/m²) every 3 weeks. However, an esophageal narrowing appeared as a consequence of tumor invasion. Therefore, an esophageal stent had been placed five months before his current admission to the hospital.

Following admission to the emergency department, his blood pressure was 89/46 mmHg, pulse rate was 119 beats per minute, regular respirations rate was 30/min, oxygen saturation was 96% on 3 L/min oxygen delivered by nasal cannula, and body temperature was 36.0°C. The patient was 158 cm tall and weighed 35 kg. No other abnormalities were found on a physical examination. Laboratory test results revealed a white blood cell count of 12,600 cells/μL, hemoglobin level of 6.5 g/dL, platelet count of 435,000/μL, international normalized ratio of prothrombin time of 1.26 (63.2%), D-dimer level of 1.2 μg/mL, and C-reactive protein level of 4.74 mg/dL. An arterial blood gas analysis revealed a pH of 7.442, partial oxygen pressure of 207 mmHg, and partial carbon dioxide pressure of 30.7 mmHg on 3 L oxygen. Aorto-esophageal fistula was suspected based on computed tomography (CT) findings (Fig. 2). Therefore, TEVAR was performed under general anesthesia because invasive surgery was considered difficult. A 10-cm-long (26 mm in diameter) conformable Gore Tag (CTAG) endoprosthesis

was inserted into zone 2 of the aortic arch; this successfully controlled the hemorrhaging, thus saving the patient's life.

On day 14 after admission, CT with oral gastrografin revealed leakage of contrast into the mediastinum. On day 28, the esophageal stent was reinserted. However, on the following day, the patient experienced a high fever with an inflammatory response, as evidenced by laboratory findings; therefore, the administration of tazobactam/piperacillin was initiated. On day 34, mediastinal air was still observed on CT, and oral feeding was considered impossible. On day 42, a gastrostomy was created through which enteral feeding was initiated several days later. Subsequently, the patient's condition remained stable, and he became well enough for discharge.

However, 63 days after the initial admission, the fever recurred with hematemesis and melena. CT revealed the recurrence of aorto-esophageal fistula and its consolidation in both lungs. Re-implantation of an intravascular stent was considered unfeasible, and the patient died the following day.

Discussion

In Japan, permanent indwelling of esophageal stents for stenosis due to malignancy has been approved and is indi-

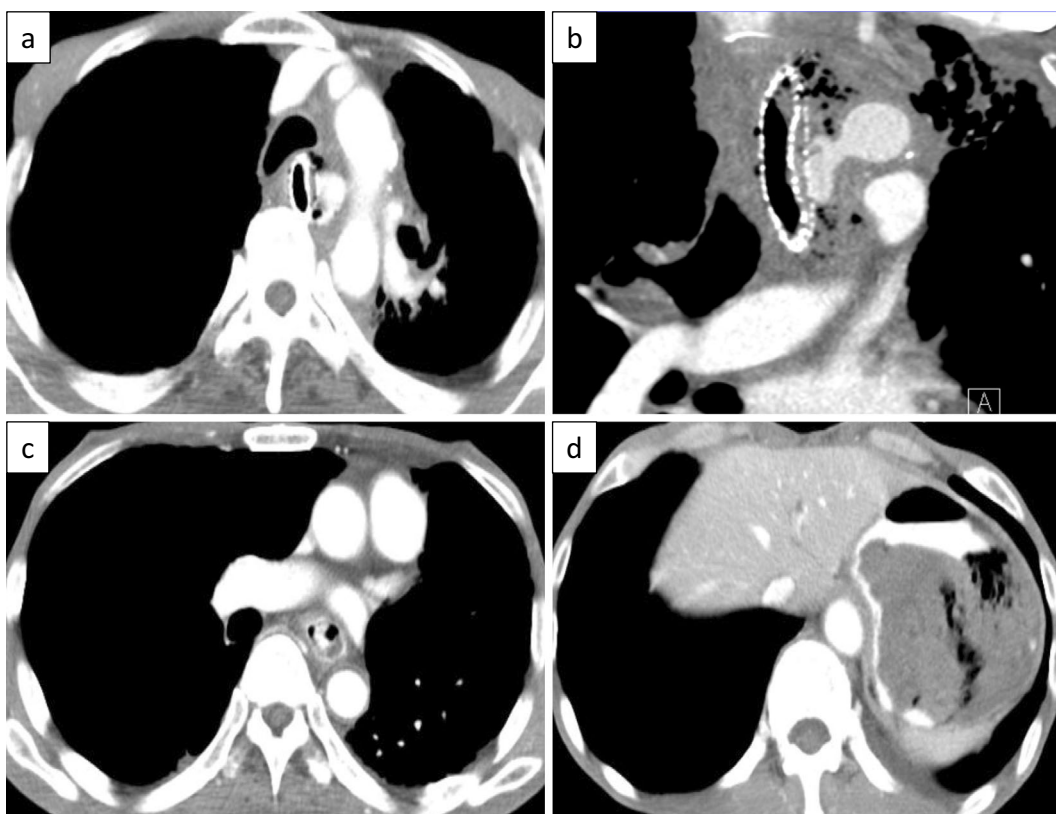


Figure 2. a-d: Leakage of the contrast agent to the mediastinum is seen, and contrast agent leakage is seen also in the stomach from the esophagus.

cated for very elderly patients and those with a poor performance status. Prompt symptomatic improvement is expected following stent placement. However, esophageal stent placement for stenosis secondary to recurrent cancer or cancer radiotherapy can result in serious complications. Indeed, in a previous study, 3 of 13 patients had esophageal perforation, 6 had mediastinitis, and 7 died of treatment-related pulmonary complications (3).

Aorto-esophageal fistula still has a high mortality rate, for which preoperative hemodynamics and mediastinitis are significant prognostic factors (4). Patients treated soon after the onset of aorto-esophageal fistula with prosthetic reconstruction of the aorta and esophagectomy with curative intent are more likely to survive than those with delayed treatment. However, a given patient's condition and degree of risk may prevent the use of such an invasive approach.

TEVAR is a minimally-invasive approach for stabilizing the hemodynamics and has been shown to have an acute hemostatic effect (1). Nonetheless, TEVAR alone is insufficient for controlling infection (5, 6). Esophagectomy with debridement of infected tissues and omentoplasty to fill the defect are considered suitable (7, 8). In a previous study, TEVAR alone was used without further treatment to address infection. Similar to the patient in the present case, it was reported that most patients died because of infection or re-penetration, which itself is believed to be caused by infection (9). Therefore, to achieve a long-term survival, the early removal of the esophagus, debridement, revascularization,

and omentoplasty are necessary (10).

In our patient, in whom an aorto-esophageal fistula occurred as a consequence of lung cancer treatment, TEVAR was a life-saving intervention. The fistula in this case was believed to have resulted from the esophageal stent placed at the irradiation site, as well as from the cancer recurrence. Generally, secondary irradiation to the same site and stent placement at the irradiation site should be avoided. However, we consulted with radiologists regarding the risks associated with secondary irradiation. Because the main irradiation fields for left upper lobe lung cancer did not overlap prior fields for left lower lobe lung cancer, we considered secondary irradiation to be acceptable as therapy in our patient. Furthermore, this patient had advanced lung cancer, and because he strongly desired even minimal oral feeding, not wanting to receive gastrostomy, we selected stent placement. Because of this treatment, this patient was able to continue oral feeding until day 34 of admission. However, as advances in cancer therapy prolong the life of many patients with advanced lung cancer, we must more carefully consider the indications for secondary irradiation and stent placement at the irradiation site more carefully.

TEVAR may be the only life-saving treatment available for patients who are unable to undergo curative surgery, including those with advanced cancer. Notably, our patient survived for approximately two months after TEVAR was performed. However, because the invasive approaches required to control infection were considered unfeasible, the

therapeutic effect was only temporary. Further studies investigating the indication for TEVAR are necessary.

The authors state that they have no Conflict of Interest (COI).

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