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# A case of separation surgery with drainage tube-less (DRESS) esophagostomy for advanced cancer with a respiratory fistula

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## ABSTRACT

**INTRODUCTION:** An esophagorespiratory fistula (ERF) can cause severe pneumonia or a lung abscess which progresses to life-threatening sepsis. A case of a patient with esophageal cancer and an esophagopulmonary fistula (EPF) who underwent separation surgery with drainage tube-less (DRESS) esophagostomy and was promptly started on definitive chemoradiotherapy (CRT) is reported.

**PRESENTATION OF CASE:** A 79-year-old man visited a clinic with a month-long history of dysphagia. Esophageal cancer at the middle thoracic esophagus was detected, and invasion of the left main bronchus and lower lobe of the right lung was seen on contrast-enhanced computed tomography (CT). Three weeks later, the patient was transferred to our hospital. CT showed a lung abscess in the lower lobe of the right lung that continued into the adjacent esophageal cancer. Due to the EPF, the patient underwent emergency surgery that consisted of esophageal separation surgery and double bilateral esophagostomy and enterostomy. Definitive CRT for the esophageal cancer was started from postoperative day 25. At six-month follow-up, the patient achieved relapse-free survival.

**DISCUSSION:** Separation surgery with a DRESS esophagostomy provides good control of inflammation because of division of the respiratory tract from the alimentary tract, which allows prompt initiation of CRT. Alternatively, a DRESS esophagostomy allows patients to be free from any tube trouble.

**CONCLUSION:** Separation surgery with a DRESS esophagostomy for an ERF is a promising method to improve patient quality of life that is less invasive, controls inflammation, and facilitates subsequent definitive CRT.

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## 1. Introduction

Once esophagorespiratory fistulas (ERF) including esophagopulmonary fistulas (EPF) occur in patients with esophageal cancer with invasion to adjacent organs, saliva and food flow into the respiratory tract through the fistula and can cause fever, cough, and dyspnea; they frequently become refractory and cause severe illness [1,6]. An esophageal covered expandable metallic stent (EMS) is a less-invasive treatment with a high rate of fistula closure [3,6]. However, since a high incidence of complications such as perforation and bleeding was noted in a previous report, the indication for stent placement should be carefully considered [3,4,9].

**Abbreviations:** DRESS, drainage tube-less; ERF, esophagorespiratory fistula; EPF, esophagopulmonary fistula; CRT, chemoradiotherapy; CT, computed tomography; EMS, expandable metallic stent; PET-CT, positron emission tomography-CT; CDDP, cisplatin; 5-FU, 5-fluorouracil.

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An esophageal cancer patient with an EPF who underwent separation surgery with drainage tube-less (DRESS) esophagostomy and whose inflammation from the EPF could be effectively controlled, which facilitated the prompt administration of definitive chemoradiotherapy (CRT), is reported.

This case report has been reported in line with the SCARE criteria [14].

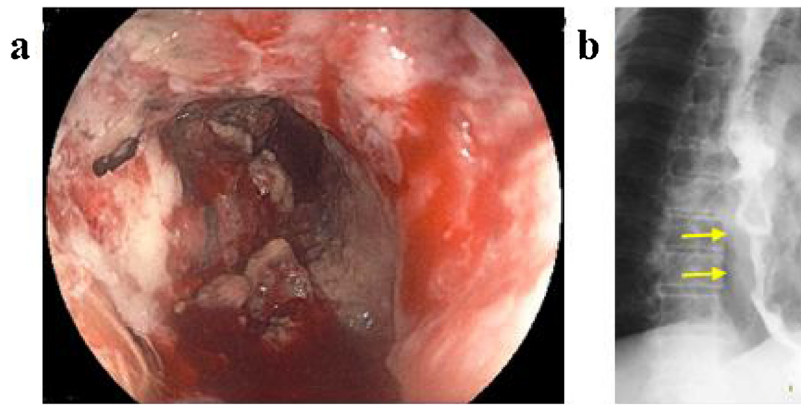
## 2. Presentation of case

A 79-year-old man visited a clinic with a month-long history of dysphagia. Esophageal cancer at the middle thoracic esophagus was seen on endoscopic examination and esophagography (Fig. 1a, b). On contrast-enhanced computed tomography (CT), the esophageal cancer seemed to invade the left main bronchus and the lower lobe of the right lung. There were metastases to regional lymph nodes, but there was no evidence of distant organ metastases.

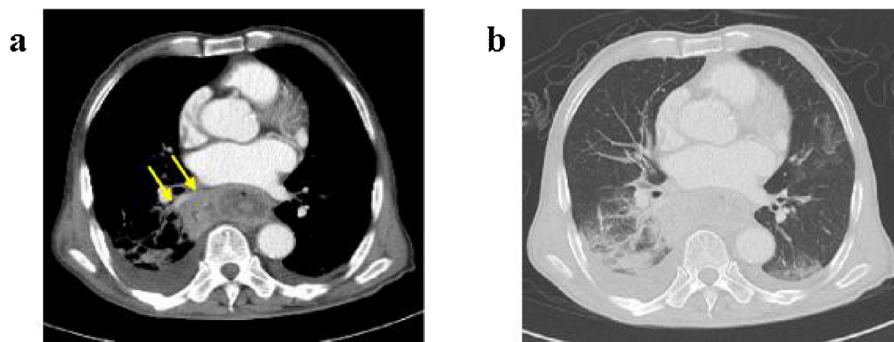
Three weeks later, the patient was transferred to our hospital and was examined in detail. Though fever and respiratory symptoms were not observed, positron emission tomography (PET)-CT and urgent CT detected a lung abscess in the lower lobe of the right

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**Fig. 1.** Endoscopic examination and Esophagography.  
 a, Endoscopic examination shows esophageal cancer with stenosis.  
 b, Esophagography (arrow) show esophageal cancer at the middle thoracic esophagus.



**Fig. 2.** Urgent computed tomography (CT) on the day of an esophagopulmonary fistula (EPF).  
 a, CT shows a lung abscess (arrow) in the lower lobe of the right lung that continues into adjacent esophageal cancer.  
 b, CT shows infiltrative shadows in the peripheral lung field and a pleural effusion.

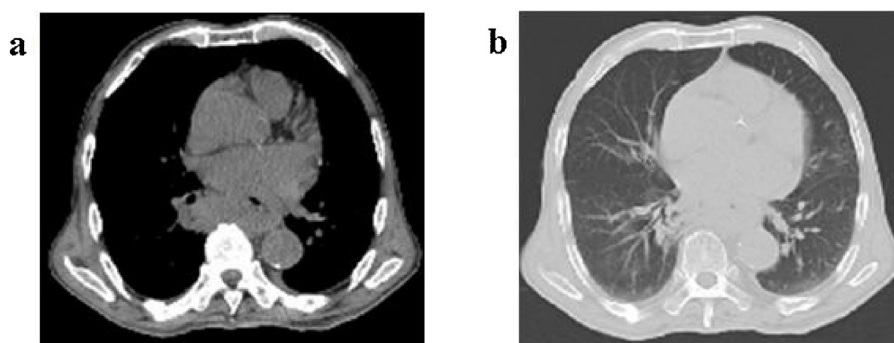
lung that continued into the adjacent esophageal cancer, infiltrative shadows in the peripheral lung field, and a pleural effusion (Fig. 2a, b). Blood tests showed an increased white blood cell (WBC) count of 9600/mm<sup>2</sup> and a C-reactive protein of 13.43 mg/dL (Table 1). Due to the presence of the EPF, the patient underwent emergency surgery that consisted of esophageal separation surgery and double bilateral esophagostomy and enterostomy. Antibiotic drug therapy for pneumonia and lung abscess achieved a favorable outcome (Fig. 3a, b).

Definitive CRT for the esophageal cancer was started from postoperative day 25. Radiotherapy was performed with a total dose of 46.8 Gy. Chemotherapy consisted of cisplatin (CDDP) and 5-fluorouracil (5-FU) therapy in 2 courses. Radiotherapy could not be

completed because of sepsis due to aspiration pneumonia. The aspiration pneumonia improved with intensive treatment. At 90 days after surgery, the patient was discharged from the hospital. At six-month follow-up, the patient had achieved relapse-free survival and is currently symptom-free.

### 2.1. Separation surgery with DRESS esophagostomy

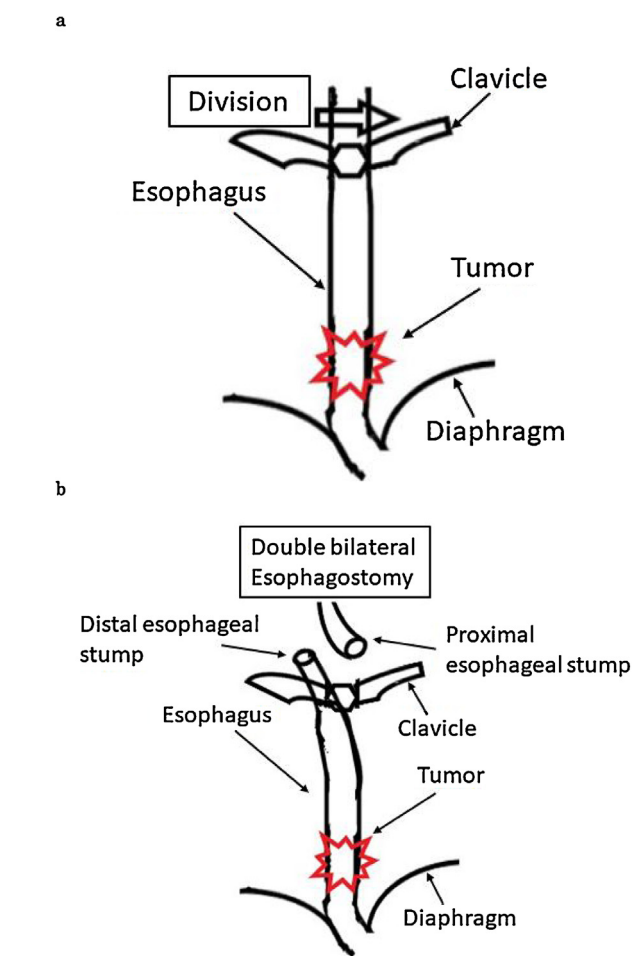
A bilateral neck diagonal incision was made. In the left side of the neck, the cervical esophagus was isolated and retracted by taping. On the right side neck, the cervical esophagus was isolated in the same way as on the left side and isolated by taping from the left side to the right side. The cervical esophagus was divided using a



**Fig. 3.** Computed tomography (CT) at postoperative 3 weeks.  
 a, CT shows that the lung abscess have decreased compared to preoperative CT.  
 b, CT shows that infiltrative shadows have disappeared.

**Table 1**  
Laboratory data on the day of an esophagopulmonary fistula (EPF). Increased white blood cell (WBC) count of 9600/mm<sup>2</sup> and C-reactive protein of 13.43 mg/dL are seen.

Laboratory result		
White blood cell count	9600	/mm <sup>2</sup>
Red blood cell count	3,380,000	/mm <sup>2</sup>
Hemoglobin	10.3	g/dl
Hematocrit	29.7	%
Blood platelet count	190,000	/mm <sup>2</sup>
Total protein	5.55	g/dl
Albumin	2.02	g/dl
Blood urea nitrogen	10.8	mg/dl
Creatinine	0.67	mg/dl
Total bilirubin	1.2	mg/dl
Aspartate aminotransferase	25	U/l
Alanine aminotransferase	15	U/l
Lactate dehydrogenase	122	U/l
Alkaline phosphatase	151	U/l
Sodium	125	mmol/l
Potassium	4.0	mmol/l
Chlorine	95	mmol/l
C-reactive protein	13.43	mg/dl
Prothrombin time (%)	65.2	%
Activated partial thromboplastin time	35.3	s



**Fig. 4.** Scheme of the Separation surgery.  
a, The cervical esophagus is divided using a linear stapler.  
b, The proximal esophageal stump is pulled out to the left side neck, and the distal esophageal stump is pulled out to the right supraclavicular region.

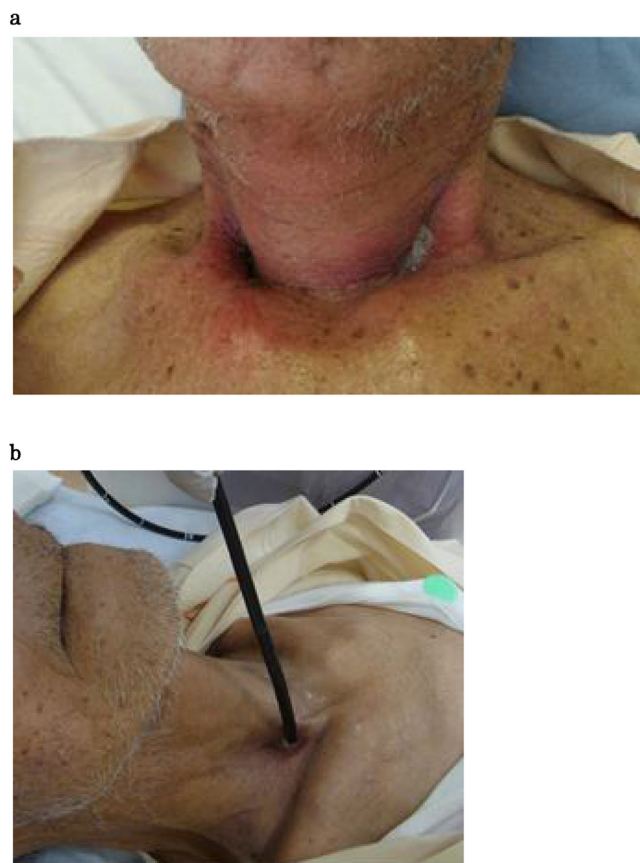
linear stapler (Fig. 4a), and the distal esophageal stump was pulled out to the right supraclavicular region. To extend the region of the esophagostomy, the sternal head of sternocleidomastoid muscle was resected, and a skin flap was made around the region of the

esophagostomy. The esophageal wall was fixed with skin all-round. In the same way, the proximal esophageal stump was pulled out to the left side neck for the esophagostomy (Figs. 4b and 5a), and then enterostomy was performed by laparotomy.

**3. Discussion**

The rate of ERFs in esophageal cancer patients with invasion to adjacent organs was reported to be approximately 5–10%, and it increased to 10–30% during or after CRT in the previous reports [1,6]. An ERF invades the trachea, bronchus, or lung, and saliva and food flow into the respiratory tract continuously through the fistula, so that severe pneumonia or lung abscess can develop. The main symptoms of ERFs are fever, cough, and dyspnea. These patients often have stenosis of the esophagus and/or dysphagia, which cause malnutrition and a poor performance status. Therefore, pneumonia due to an ERF that progresses to sepsis is a life-threatening condition. Thus, ERFs should be diagnosed immediately, and rapid primary treatment is needed.

Alternatively, the treatment for the esophageal cancer in patients with an ERF should be considered concurrently with the treatment for pneumonia. Patients with an ERF have evidence of invasion into adjacent organs and a poor prognosis [2]. Definitive CRT for patients with invasion into adjacent organs produced a complete response in approximately 20–30% and 5-year survival of approximately 7–14% in the previous reports [2,10,11]. It was reported that the median survival period of the patients with ERFs who underwent palliative care ranged from 1 to 6 weeks due to their poor condition [1,6]. The treatment strategy for an ERF is to



**Fig. 5.** Drainage tube-less (DRESS) esophagostomy.  
a, Double bilateral esophagostomy of a 79-year-old man.  
b, Endoscopic investigations through the DRESS esophagostomy to evaluate esophageal cancer.



first control the respiratory symptoms and inflammation, giving priority to immediate life-saving treatment. Second, CRT should be promptly performed in situations of controlled inflammation to improve the prognosis.

Various treatments for ERFs have been reported previously, such as esophageal covered EMS. Placement of a covered EMS in esophageal cancer patients with ERFs makes it possible to seal off the fistula with a high rate of success and contribute to less-invasive treatment [3,6]. However, fistula regression was reported in 35% of patients [3,4], and CRT during stent placement causes severe complications, such as bleeding or worsening pneumonia due to spread of the fistula; treatment-related death from CRT during stent placement was reported in 21% of patients [9]. Considering the high severe complication rate of this treatment, the indication for CRT during stent placement should be carefully considered after obtaining informed consent.

Esophagectomy with combined resection of invaded organs or bypass surgery is reported to prolong survival [5,12,13], but its high morbidity is also well known. Thus, postoperative complications might delay definitive CRT, and such surgeries do not necessarily benefit such patients.

Considering control of inflammation and initiation of CRT promptly as the primary option, less invasive treatments should be selected. Esophageal separation surgery provides good control of inflammation because of division of the respiratory tract from the alimentary tract certainly. Additionally, it is less invasive than esophageal bypass surgery in that the operative procedure involves only the cervical region, and no esophageal anastomosis is required, which allows prompt initiation of CRT. In the present case, there were no complications related to the esophagostomy, but complications such as stomal falling and subcutaneous leakage have been generally reported. The skin flap around the esophagostomy should be extensive, and the esophageal wall should be sutured with skin in a tensionless manner to prevent stomal falling.

In many cases of esophageal surgeries, an external esophagostomy is made with a drainage tube. An esophagostomy is placed in the right supraclavicular region or the left hypochondriac region, and indwelling drainage tubes were inserted into the esophagus to avoid creating a closed space at the oral and anal esophagus separated due to the esophageal cancer in the previous reports [2,7,8]. Therefore, drainage tubes sometimes cause trouble and affect the quality of life of patients after esophageal surgery. Meanwhile, esophagocutaneostomy can be managed easily without a drainage tube because of little fluid discharge. Therefore, our DRESS esophagostomy might improve patient's quality of life. Additionally, DRESS esophagostomy is useful for endoscopic investigations to evaluate esophageal cancer or the fistula (Fig. 5b).

#### 4. Conclusion

Separation surgery with a DRESS esophagostomy for an ERF is a promising method to improve patient's quality of life in advanced stage esophageal cancer with less invasiveness and with control of inflammation, facilitating subsequent definitive CRT.

#### Conflict of interest

The authors have no financial and personal relationships with other people or organisations, and no potential conflicts of interest to declare.

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#### Ethical approval

This research study was approved by the ethics committee of Miyazaki University Hospital. The name of the relevant ethics committee is the ethics committee of Miyazaki University Hospital, the reference number of this research is O-0182, and the name of this research is study on ingenuity of esophageal cancer surgery technique.

#### Consent

The authors obtained written informed consent from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review on request. Patients' names, initials, or hospital numbers was not used in our article and images of patients.

#### Author contribution

Yukinori Tanoue: Participated substantially in conception, design, and execution of the paper and in the interpretation of data; also participated substantially in the drafting and editing of the manuscript.

Shinsuke Takeno: Participated substantially in conception, design, and execution of the paper and in the interpretation of data; also participated substantially in the drafting and editing of the manuscript.

Fumiaki Kawano: Participated substantially in execution of the paper and in the interpretation of data.

Kousei Tashiro: Participated substantially in execution of the paper and in the interpretation of data.

Rouko Hamada: Participated substantially in design, and execution of the paper and in the interpretation of data.

Yasuyuki Miyazaki: Participated substantially in design, and execution of the paper and in the interpretation of data.

Atsushi Nanashima: Participated substantially in conception of the paper; also participated substantially in the editing of the manuscript.

#### Guarantor

Yukinori Tanoue, M.D., Shinsuke Takeno, M.D.

#### References

- [1] M. Muto, A. Ohtsu, S. Miyamoto, K. Muro, N. Boku, S. Ishikura, et al., Concurrent chemoradiotherapy for esophageal carcinoma patients with malignant fistulae, *Cancer* 8 (October (8)) (1999) 1406–1413.
- [2] J. Hihara, Y. Hamai, M. Emi, Y. Aoki, J. Taomoto, Y. Miyata, et al., Esophageal bypass operation prior to definitive chemoradiotherapy in advanced esophageal cancer with tracheobronchial invasion, *Ann. Thorac. Surg.* 97 (January (1)) (2014) 290–295.
- [3] J.H. Shin, H.Y. Song, G.Y. Ko, J.O. Lim, H.K. Yoon, K.B. Sung, Esophagorespiratory fistula: long-term results of palliative treatment with covered expandable metallic stents in 61 patients, *Radiology* 232 (July (1)) (2004) 252–259.
- [4] M. Yakami, M. Mitsumori, H. Sai, Y. Nagata, M. Hiraoka, Y. Nishimura, Development of severe complications caused by stent placement followed by definitive radiation therapy for T4 esophageal cancer, *Int. J. Clin. Oncol.* 8 (December (6)) (2003) 395–398.
- [5] T. Matsubara, M. Ueda, N. Kokudo, T. Takahashi, T. Muto, A. Yanagisawa, Role of esophagectomy in treatment of esophageal carcinoma with clinical evidence of adjacent organ invasion, *World J. Surg.* 25 (March (3)) (2001) 279–284.
- [6] A. Turkyilmaz, Y. Aydin, A. Eroglu, Y. Bilen, N. Karaoglanoglu, Palliative management of esophagorespiratory fistula in esophageal malignancy, *Surg. Laparosc. Endosc. Percutan. Tech.* 19 (October (5)) (2009) 364–367.
- [7] Y. Seto, K. Yamada, T. Fukuda, N. Hosoi, R. Takebayashi, K. Chin, et al., Esophageal bypass using a gastric tube and a cardiostomy for malignant esophagorespiratory fistula, *Am. J. Surg.* 193 (June (6)) (2007) 792–793.
- [8] T. Hirai, Y. Yamashita, H. Mukaida, K. Kawano, T. Toge, M. Niimoto, et al., Bypass operation for advanced esophageal cancer—an analysis of 93 cases, *Jpn. J. Surg.* 19 (March (2)) (1989) 182–188.

- [9] Y. Nishimura, K. Nagata, S. Katano, S. Hirota, K. Nakamura, F. Higuchi, et al., Severe complications in advanced esophageal cancer treated with radiotherapy after intubation of esophageal stents: a questionnaire survey of the Japanese Society for Esophageal Diseases, *Int. J. Radiat. Oncol. Biol. Phys.* 56 (August (5)) (2003) 1327–1332.
- [10] T. Makino, Y. Doki, Treatment of T4 esophageal cancer: definitive chemo-radiotherapy vs chemo-radiotherapy followed by surgery, *Ann. Thorac. Cardiovasc. Surg.* 17 (3) (2011) 221–228.
- [11] H. Fujita, S. Sueyoshi, T. Tanaka, Y. Tanaka, S. Matono, N. Mori, et al., Esophagectomy: is it necessary after chemoradiotherapy for a locally advanced T4 esophageal cancer? Prospective nonrandomized trial comparing chemoradiotherapy with surgery versus without surgery, *World J. Surg.* 29 (January (1)) (2005) 25–30.
- [12] T. Matsubara, M. Ueda, T. Nakajima, K. Nakagawa, T. Yamashita, N. Horikoshi, et al., Can esophagectomy cure cancer of the thoracic esophagus involving major airways? *Ann. Thorac. Surg.* 59 (January (1)) (1995) 173–177.
- [13] I. Yamana, S. Takeno, R. Shibata, H. Shiwaku, K. Maki, T. Hashimoto, et al., Esophagectomy and combined lobectomy with a covering latissimus dorsi muscle flap for an esophagopulmonary fistula due to advanced esophageal cancer: report of two cases, *Esophagus* 11 (2014) 277–281.
- [14] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, for the SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (October) (2016) 180–186.

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