



Palliation of malignant esophageal stent esophagorespiratory fistula with rigid bronchoscope and Y silicone trachea stent: experience with seven patients

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Background: Malignant esophageal stent esophagorespiratory fistula (ERF) is an abnormal communication between esophagus and airway among advanced tumor patients with indwelling esophageal stent, which is devastating and life-threatening. This study aims to provide a new feasible treatment scheme for malignant esophageal stent ERF and report its potential advantage compared with double stenting, which was recommended by European Society of Gastrointestinal Endoscopy Guideline.

Methods: We retrospectively analyzed the medical data of malignant esophageal stent ERF patients between January 2018 to May 2023 at the First Affiliated Hospital of Guangzhou Medical University and divided them into two groups. Group 1 consisted of patients treated with rigid bronchoscopy to remove the esophageal stent and implant Y silicone trachea stent, while group 2 consisted of patients treated with additional airway stenting without removing the esophageal stent. Demographic parameters, disease diagnoses and treatment, radiological findings before and after the intervention, and complications caused by the stents were obtained and analyzed with chi-squared, Mann-Whitney *U*, independent-samples *t*-tests, Kaplan-Meier methods, and log-rank test.

Results: Ten patients (seven patients in group 1 and three in group 2) were included. No procedure complications occurred in both groups. The mean Karnofsky Performance Score after the procedure significantly improved compared to the pre-procedure (57.14 vs. 77.14, $P=0.001$) in group 1, while decreased in group 2 (50 vs. 40, $P=0.026$). The control of pneumonia in group 1 patients is better than that in group 2. There was significant improvement in the degree of dysphagia after the procedure (3.86 vs. 2.43, $P=0.002$) in group 1, while no improvement was found in group 2 (4.00 vs. 3.33, $P=0.423$). The mean survival of group 1 was significantly longer group 2 (381.00 vs. 80.33 days, $P<0.001$, log-rank test). No patient needed stent repositioning due to migration in both groups. Cause of death in the group 1 included disease progression, novel coronavirus pneumonia, massive hemoptysis, and respiratory insufficiency, while group 2 included severe pneumonia and disease progression. No death was directly attributed to the procedure in both groups.

Conclusions: Removing the esophageal stent and implanting Y silicone trachea stent through a rigid bronchoscopy is a safe and feasible treatment for malignant esophageal stent ERF. This procedure can

effectively seal the fistula, prevent from recurrent aspiration pneumonia, improve the quality of life, and prolong the survival time.

Keywords: Malignant esophageal stent esophagorespiratory fistula (malignant esophageal stent ERF); double stent; rigid bronchoscope; Y silicone trachea stent

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Introduction

Malignant dysphagia and esophagorespiratory fistula (ERF) are common symptoms of esophageal cancer and invasion of esophagus and trachea by other primary tumors, such as lung cancer and gastric cancer (1). Malignant ERF is an abnormal communication between esophagus and airway, which is devastating and life-threatening (1). Esophageal stenting is an effective way to palliate malignant dysphagia and seal malignant ERF (1). However, with the increase of the stenting duration and the impact of radiation therapy and chemotherapy, the likelihood of malignant esophageal stent ERF increases (2-5). Malignant esophageal stent ERF is defined as patients who develop ERF after esophageal stent placement (2).

The management of malignant esophageal stent ERF

is urgent and challenging. These patients generally have advanced tumor stage, poor physical health, and may not withstand surgery (6). Without appropriate treatment, these patients are likely to die from lung infection resulting from chronic aspiration through fistula (6). Currently, endoscopic treatment is regarded as the preferred option for the management of malignant esophageal stent ERF (6). However, the reported endoscopic treatment methods for malignant esophageal stent ERF are basically based on the treatment for ERF, such as additional esophageal and/or tracheal stents, which may increase pressure on the affected tissue, ultimately creating a larger fistula (2,7-9).

In fact, the endoscopic treatment for ERF and malignant esophageal stent ERF should be quite different. Esophageal stent was a useful tool for sealing fistulas in ERF treatment, but for malignant esophageal stent ERF treatment, they are a risk factor for fistula deterioration and stumbling block to further endoscopic treatment and ideal therapeutic effect.

In this study, we provided a new feasible treatment scheme for malignant esophageal stent ERF, with a rigid bronchoscopy to remove the indwelling esophageal stent and implant a Y silicone trachea stent. Besides, we compare our treatment scheme with double stent treatment to prove the feasibility and advantage. We present this article in accordance with the STROBE reporting checklist (available at <https://jtd.amegroups.com/article/view/10.21037/jtd-23-1298/rc>).

Methods

Patients

We retrospectively analyzed the medical data of malignant esophageal stent ERF between January 2018 to May 2023 at the First Affiliated Hospital of Guangzhou Medical University. Based on different treatment procedure, these patients were divided into two groups. Group 1 consisted

Highlight box

Key findings

- Removing the esophageal stent and implanting Y silicone trachea stent through a rigid bronchoscopy is a safe and feasible treatment for malignant esophageal stent esophagorespiratory fistula (ERF). This procedure can effectively seal the fistula, prevent from recurrent aspiration pneumonia, improve the quality of life, and prolong the survival time.

What is known and what is new?

- Additional esophageal and/or tracheal stents was considered as a treatment option for malignant esophageal stent ERF. However, additional stents may exert increased pressure on the affected tissue, potentially leading to the enlargement of the fistula.
- This study provided a new feasible treatment scheme for malignant esophageal stent ERF.

What is the implication, and what should change now?

- Removing the esophageal stent and implanting Y silicone trachea stent through a rigid bronchoscopy is a safe and feasible treatment for malignant esophageal stent ERF.

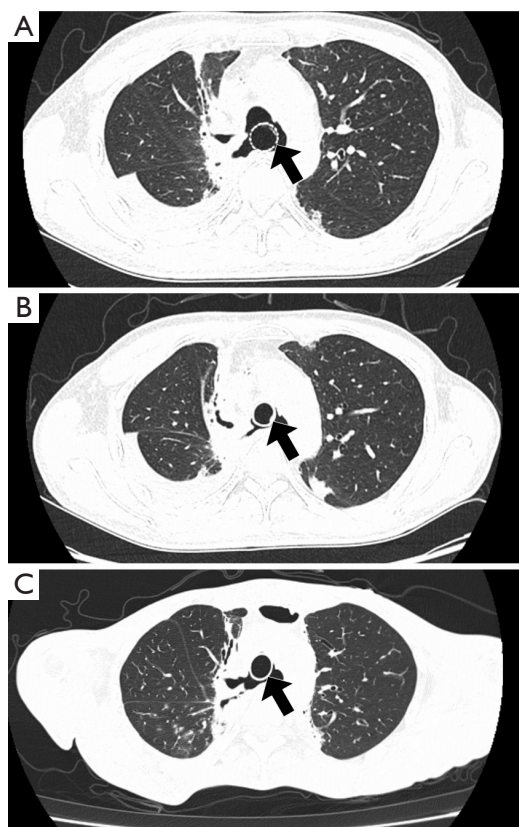


Figure 1 Computed tomography of patient 5 before and after the procedure. (A) Communication of trachea and esophagus with indwelling esophageal stent (black arrow); (B) 7 days after the procedure (black arrow points to the Y silicone stent); (C) 20 months after the procedure (black arrow points to the Y silicone stent).

of patients treated with rigid bronchoscopy to remove the esophageal stent and implant Y silicone trachea stent, while group 2 was patients treated with additional airway stenting without removing the esophageal stent. All patients gave their informed consent following detailed explanation of the risks of the procedure and the possible therapeutic alternatives. Demographic parameters, disease diagnoses, treatment details, pre- and post-procedure complications, radiological and bronchoscopy findings were obtained from patient files.

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013) (10). The study was approved by the Ethics Committee of the First Affiliated Hospital of Guangzhou Medical University (approval No. 202050). Individual consent for this

retrospective analysis was waived.

Endoscopic procedure

Prior to the endoscopic procedure, a comprehensive evaluation was conducted on all patients, including laboratory tests, radiological examinations, and bronchoscopy findings. Both flexible bronchoscopy and computed tomography (CT) scans were utilized to assess airway anatomy and formulate an appropriate treatment plan (Figures 1,2). Treatment strategies were decided by multidisciplinary team, including the respiratory intervention department, gastrointestinal endoscopy department, imaging department, and digestive surgery department. The inclusion criteria were as follows: (I) continuous indwelling esophageal self-expanding metal stent (SEMS); (II) trachea, carina, or left/right main bronchial fistula; (III) with or without trachea, main bronchial stenosis. The exclusion criteria were as follows: (I) rigid bronchoscopy cannot be inserted in airway (e.g., abnormal anatomy of the airway); (II) general anesthesia cannot be tolerated.

During the surgical procedure, the patient was positioned in a supine posture. Continuous monitoring of vital signs, including blood pressure, heart rate, electrocardiogram, and oxygen saturation, was conducted. After administering general anesthesia with mask oxygen inhalation, tracheal intubation with an endotracheal tube size less than 7.0 was performed using a video laryngoscope and a tracheal catheter. We used endotracheal tube size less than 7.0 to ensure sufficient space for the rigid bronchoscope to enter the esophagus. Once proper placement of the tracheal tube was confirmed, mechanical ventilation was initiated using an anesthesia machine for all patients. After that, we inserted the rigid bronchoscopy into the esophagus with necessary balloon expansion and thermal ablation to remove the esophageal metal stent (Video 1). The incarcerated esophageal stent was folded and rotated by rigid forceps for removing (Video 2). Once the esophageal stent was removed, we removed the trachea intubation and inserted the rigid bronchoscope in the trachea with ventilation maintained via the side port of the rigid bronchoscope. Then, the Y silicone stent was deployed and positioned in the airway to fully cover the fistula (Video 3). The edge of the stent extended at least 15 mm beyond the fistula edge. If the fistula was too large to be covered or the edge of the Y silicone stent can only barely cover the edge of the fistula, we would combine it with a covered SEMS (Video 4). Once finishing the procedure, bronchoscopy was conducted to assess the position and patency of the stent as well as the

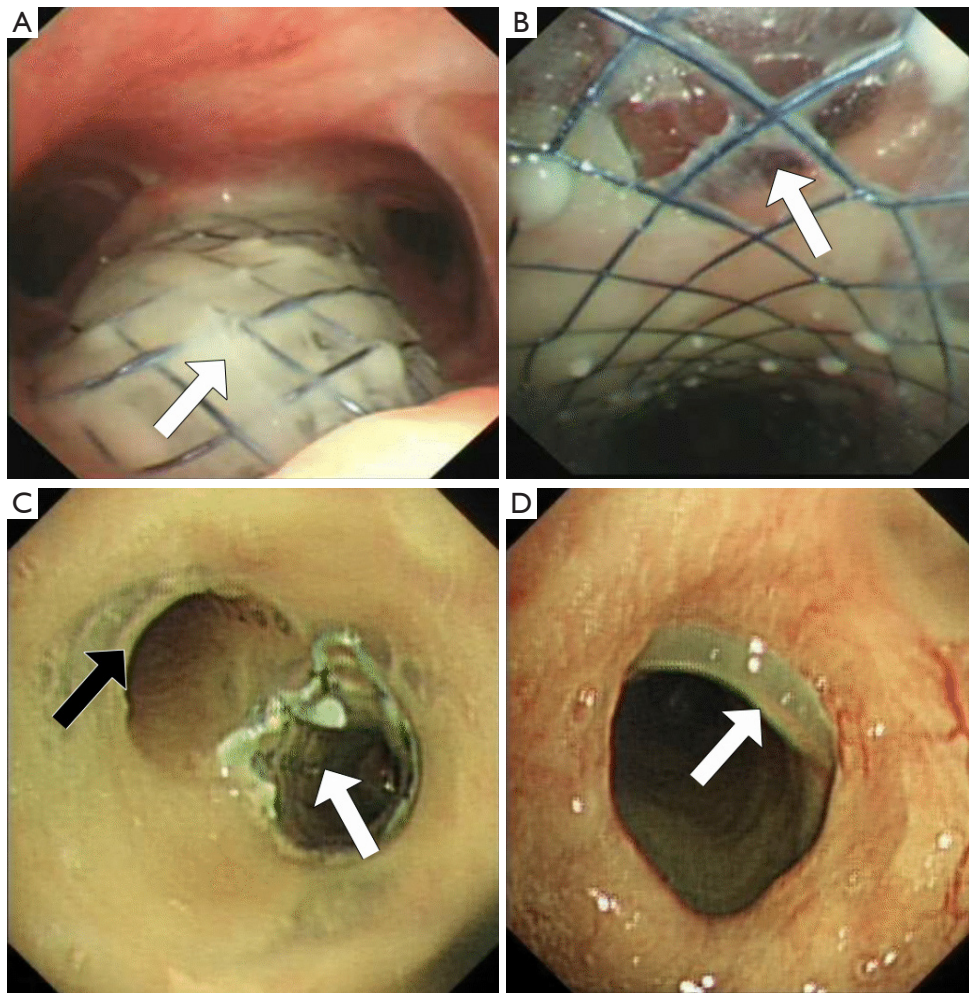
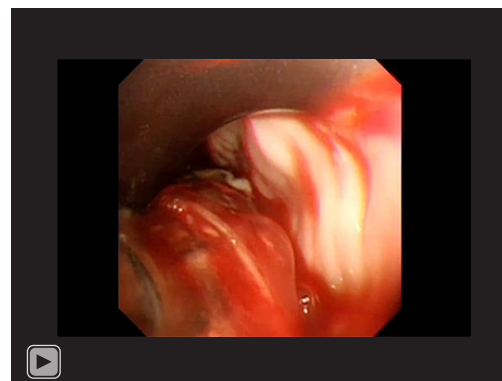


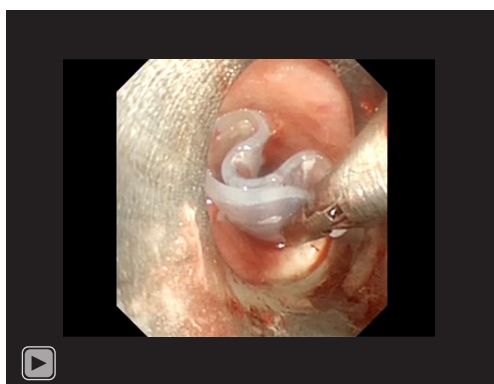
Figure 2 Bronchoscopy images of patient 5 before and after the procedure. (A) Esophageal stent (white arrow) involved carina causing significant compromise of the air lumen; (B) image of fistula (white arrow) from the esophagus side; (C) combination of Y silicone stent (black arrow) and SEMS (white arrow); (D) proximal edge of Y silicone stent (white arrow). SEMS, self-expanding metal stent.



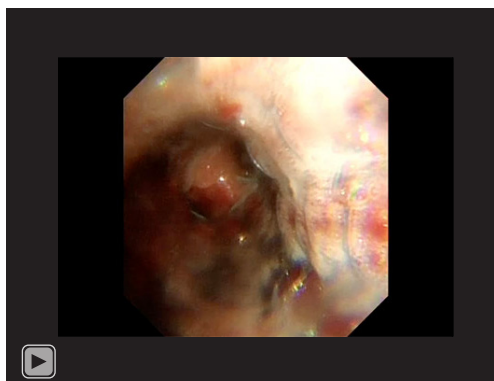
Video 1 Thermal ablation before removing the esophageal metal stent.



Video 2 Removing the esophageal metal stent.



Video 3 Placing the Y silicone stent.



Video 4 Bronchoscope of patient 5 19 months after the procedure.



Video 5 Using methylene blue to examine the stent sealing effect on the fistula.

integrity of the fistula seal. CT scan was conducted after the patient resuscitated and stabilized. The duration of the operation, from the initial of mask oxygen inhalation to the

stent placement was recorded.

Postoperative management

Patients can try eating or drinking 6 hours after anesthesia of the procedure. We suggest patients drink water first to check for irritating cough. If not, they can eat according to their own condition and comfort level, gradually transitioning to a semi-liquid and normal diet. If the patient suffers from severe irritating cough after the procedure, a CT scan and bronchoscopy will be arranged. If no abnormalities are seen on the CT scan and bronchoscopy, we will inject methylene blue into the esophagus to check for blue secretion overflowing at the fistula opening, inside the stent, the edges of the stent (*Video 5*). However, even if the fistula was greatly sealed and the patients have no irritating coughing during swallowing, we recommend them to undergo percutaneous endoscopic gastrostomy or jejunostomy to set up the way of enteral nutrition, to reduce the stimulation of food on the esophageal fistula during repeated swallowing and reduce the risk of further enlargement of the fistula. If the patients have a short-expected survival time, the final choice of eating way is up to the patient, to improve the end-of-life quality for patients.

Follow-up

All patients were followed up from the day they underwent the procedure. The follow-up time, as well as the survival time was defined as days from the procedure to death. Patients were followed up by bronchoscopy to evaluate the position and patency of the stent as well as the integrity of the fistula seal. Any symptoms associated with the procedure, such as expectoration difficulty, retrosternal pain, coughing, and shortness of breath, were recorded. Complications associated with stent implantation, such as migration, granulation, sputum retention, fistula enlargement, and new fistula occurrence were recorded. If granulation hyperplasia causes less than 50% lumen narrowing and have no related symptoms, we observe without intervention. Only if it exceeds 50% and has obvious symptoms, we perform interventional treatment. Sputum retention after tracheal stenting was managed using anti-infective drugs, atomization, or suction under bronchoscope. If the stent is displaced or cannot effectively cover an enlarged or new fistula, we will adjust or replace it. Chest radiographs or CT scan were used to assess the stent location and to identify any potential complications,

including pneumomediastinum, pneumothorax, and pneumonia. Quality of life was assessed by Karnofsky Performance Score (KPS) before and 30 days after stenting (11). Patients' dysphagia score was measured using Mellow-Pinkas scoring system, as follows: 0 = no dysphagia; 1 = dysphagia to normal solid food; 2 = dysphagia to soft solid food; 3 = dysphagia also with liquids; 4 = inability to swallow saliva (12).

Statistical analysis

All statistical analyses were performed using R (version 4.3.1). Comparisons between two independent groups were made using chi-squared, Mann-Whitney *U* and independent-samples *t*-tests as appropriate. Differences of $P < 0.05$ were considered statistically significant. Probabilities of surviving were calculated using the Kaplan-Meier estimator.

Results

Patient characteristics

Ten patients with malignant esophageal stent ERF were included and divided into two groups. Group 1 consisted of seven patients treated with rigid bronchoscopy to remove the esophageal stent and implant Y silicone trachea stent, while group 2 consisted of three patients treated with additional airway stenting without removing the esophageal stent (Table 1). The mean age of patients was 58.30 ± 9.78 years. All patients included were male. Seven patients had esophagus cancer, one patient had gastric cancer, and two patients had lung cancer. The details of the fistula, including the location, the size, and the cause of the fistula are shown in Table 1. Four patients were implanted with esophageal stent for esophageal stenosis, four patients were for esophageal fistula, one patient was for ERF, and one for esophagomediastinal fistula. There was no significant difference in the indwelling time of esophageal stent between group 1 and group 2 (Mann-Whitney *U* test, $P = 0.425$). The summary of the clinical characteristics for the ten enrolled patients and their detailed previous tumor-specific therapies are presented in Table 1.

Clinical outcomes

All indwelling esophageal stents in group 1 were removed successfully with a Y silicone stent implanted in the trachea.

All stents were successfully implanted in the airway in group 2. The detailed characteristics of the procedure are shown in Table 2. One patient had $16 \text{ mm} \times 13 \text{ mm} \times 13 \text{ mm}$ silicon stents, and six patients had $18 \text{ mm} \times 14 \text{ mm} \times 14 \text{ mm}$ silicon stents. The range affected by the fistulas and airway stenosis of patients 3 and 5 were too extensive to be covered by a Y silicone stent alone. Segmented silicone stent or/and SEMs were used to combine with the Y silicone stent (Figure 2C). The mean operation duration was 86.57 ± 3.87 minutes in group 1 and 37.67 ± 2.52 minutes in group 2. There was no significant difference between the operation time for fistula caused by tumor invasion and external compression (88.33 vs. 85.25 minutes, $P = 0.39$) (Figure S1). No procedure complications occurred. Only minor hemorrhage occurred during esophageal stent removal, and the bleeding ceased spontaneously. The result of bronchoscopy and CT after the procedure showed that the fistulas were completely sealed (Figures 1,2, Video 4).

Follow-up and complications

The follow-up outcomes of patients are shown in Table 3. The mean discharged time in group 1 (5.14 ± 3.63 days) was shorter than group 2 (10.67 ± 1.15 days) after the procedure ($P = 0.018$). The mean KPS after the procedure significantly improved compared to the pre-procedure (57.14 vs. 77.14 , $P = 0.001$) in group 1, while decreased in group 2 (50 vs. 40 , $P = 0.026$). No patient needed stent repositioning due to migration in both groups. Patients 3 and 4 complained of retrosternal pain after the procedure. To alleviate the pain, we replaced the Y silicone stent with a smaller size of $16 \text{ mm} \times 13 \text{ mm} \times 13 \text{ mm}$, which resulted in relief of the retrosternal pain. Three patients in group 1 received an additional SEMs during the follow up due to stenosis caused by tumor progression (patient 6) or granulation outside the edge of the Y silicone stent (patients 1 and 7). Four of 7 patients showed sputum retention in group 1, which was aspirated under bronchoscopy. The control of pneumonia in group 1 patients is better than that in group 2. All patients in group 1 showed improvement in pneumonia after the procedure (Figure 3). There was significant improvement in the degree of dysphagia after the procedure (3.86 vs. 2.43 , $P = 0.002$) in group 1, while no improvement was found in group 2 (4.00 vs. 3.33 , $P = 0.423$). The mean survival of group 1 was significantly longer than that of group 2 (381.00 vs. 80.33 days, $P < 0.001$, log-rank test; Figure 4). Cause of death in the group 1 included disease progression, novel coronavirus pneumonia, massive

Table 1 Baseline characteristics of patients

Group	Patient	Sex/age (years)	Primary tumor	Tumor stage	Pathologic pattern	Location of fistula (airway side)	Size of fistula (mm)	Cause of fistula	Indication for esophageal stenting	Esophageal stent indwelling time (days)	Previous tumor-specific therapy
1	1	M/55	Esophageal cancer	T4N1M1	SCC	Carina and left main bronchus	10×10	Tumor invasion	Esophageal stenosis	85	Chemotherapy, radiotherapy, and esophageal stent
	2	M/64	Gastric cancer	-	-	Lower trachea and left main bronchus	15×20, 12×20	External compression	Esophageal fistula	95	Esophageal stent, partial gastrectomy, and chemotherapy
	3	M/72	Esophageal cancer	T4N2M1	SCC	Upper trachea and left main bronchus	12×30	Tumor invasion	Esophageal fistula	462	Esophageal stent, chemotherapy, radiotherapy, immunotherapy, targeted therapy, and jejunostomy
	4	M/63	Esophageal cancer	T3N1M0	SCC	Middle and lower trachea	18×30	Tumor invasion	Esophageal stenosis	78	Radiotherapy, chemotherapy, and esophageal stent
	5	M/49	Lung cancer	T4N3M1c	LCC	Lower trachea and carina	20×25	External compression	Esophagorespiratory fistula	62	Radiotherapy, chemotherapy, and esophageal stent
	6	M/47	Esophageal cancer	T4N1M1	SCC	Upper and lower trachea, left main bronchus	10×10, 16×20, 15×15	External compression	Esophagomediastinal fistula	181	Esophageal stent, radiotherapy, chemotherapy, and immunotherapy
	7	M/54	Esophageal cancer	T4N1M1	SCC	Middle trachea and 5 cm above the carina	15×10	External compression	Esophageal stenosis	164	Esophageal stent, radiotherapy, chemotherapy, and immunotherapy
2	8	M/58	Esophageal cancer	T4N2M1	SCC	Left main bronchus	20×15	External compression	Esophageal stenosis	183	Esophageal stent, radiotherapy, chemotherapy
	9	M/74	Lung cancer	T3N3M1c	SCC	Middle trachea and right intermediate bronchus	20×20, 5×5	External compression	Esophageal fistula	50	Chemotherapy, and esophageal stent
	10	M/47	Esophageal cancer	T4N1M1	SCC	Lower trachea	5×10	External compression	Esophageal fistula	67	Chemotherapy, radiotherapy, and esophageal stent

Group 1: patients treated with rigid bronchoscopy to remove the esophageal stent and implant Y silicone trachea stent; Group 2: patients treated with additional airway stenting without removing the esophageal stent. Tumor stage was assessed based on the 8th American Joint Committee on Cancer/Union for International Cancer Control (AJCC/UICC) system. The size of fistula is presented as length × width. M, male; SCC, squamous cell carcinoma; LCC, large cell carcinoma.

Table 2 Characteristics of the procedure

Patient No.	Stent in trachea	Parameters of stent in trachea (mm)	Stent in bronchus	Parameters and locations of stent in bronchus (mm)	Operation duration (minutes)
1	Y silicone stent	16×13×13/40×25×5*	–	–	92
2	Y silicone stent	18×14×14/50×45×15*	–	–	81
3	Y silicone stent, segmented silicone stent	18×14×14/50×20×15*, 20×40×18 [†]	SEMS	14×40 [‡] (left main bronchus)	83
4	Y silicone stent	18×14×14/65×15×15*	–	–	90
5	Y silicone stent	18×14×14/50×35×15*	SEMS	14×40 [‡] (right main bronchus)	85
6	Y silicone stent	18×14×14/80×35×15*	–	–	87
7	Y silicone stent	18×14×14/75×20×15*	–	–	88
8	–	–	Y SEMS	14×10×10/50×10×10 [§] (left main bronchus, left upper lobar bronchus, left inferior lobar bronchus)	40
9	SEMS	16×60 [‡]	SEMS	10×30 [‡] (right intermediate bronchus)	35
10	SEMS	16×60 [‡]	SEMS	10×40 [‡] (left main bronchus)	38

*, parameters of Y silicone stent are presented as diameter of tracheal body × left limb × right limb/length of tracheal body × left limb × right limb; [†], parameters of segmented silicone stent are presented as external diameter × length × internal diameter; [‡], parameters of SEMS are presented as diameter × length; [§], parameters of Y SEMS are presented as diameter of left main bronchus limb × left upper lobar bronchus × left inferior lobar bronchus/length of left main bronchus limb × left upper lobar bronchus × left inferior lobar bronchus. SEMS, self-expandable metal stent.

Table 3 Follow-up outcome of patients

Patient number	Degree of dysphagia before stenting/after stenting	KPS before stenting/after stenting	Stent-related complications	Survival (days)	Tumor-specific therapy after stenting
1	4/3	60/80	Granulation	318	Gastrostomy, SEMS placement and replacement, chemotherapy, and radiotherapy
2	3/2	60/80	Retrosternal pain, sputum retention	575	Chemotherapy, Y silicone stent replacement
3	4/3	60/80	NA	389	Chemotherapy, radiotherapy, immunotherapy, and targeted therapy
4	4/3	50/70	Retrosternal pain	237	Gastrostomy, Y silicone stent replacement, radiotherapy, and chemotherapy
5	4/3	60/80	Granulation, sputum retention	602	Jejunostomy, radiotherapy, and chemotherapy
6	4/2	50/70	Sputum retention	154	SEMS placement, radiotherapy, and chemotherapy
7	4/1	60/80	Granulation, sputum retention	392	Y silicone stent replacement, SEMS placement, laser resection, balloon dilation, radiotherapy, chemotherapy, and immunotherapy
8	4/4	50/20	Sputum retention	19	Jejunostomy
9	4/2	50/70	Fistula occurrence	147	Targeted therapy
10	4/4	50/30	Sputum retention	75	Chemotherapy, immunotherapy, and jejunostomy

KPS, Karnofsky Performance Status; SEMS, self-expandable metal stent; NA, not available.

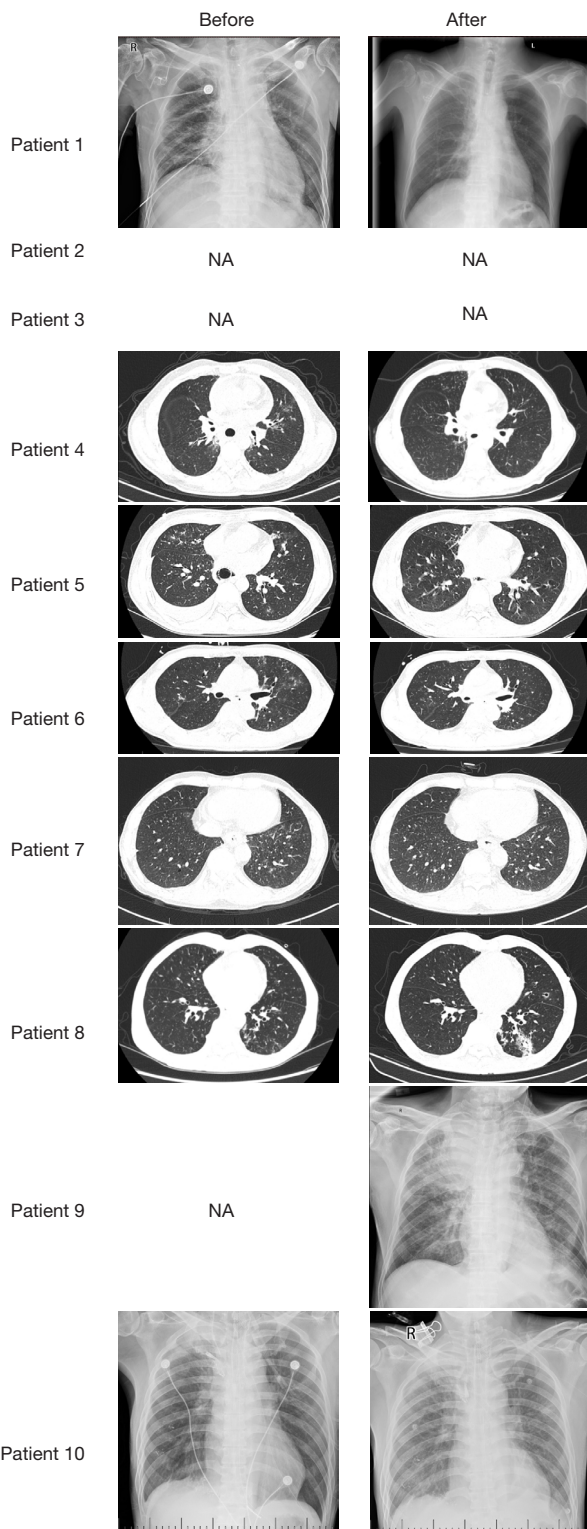


Figure 3 The comparison of preoperative and postoperative chest X-ray or CT images. NA, not available; CT, computed tomography.

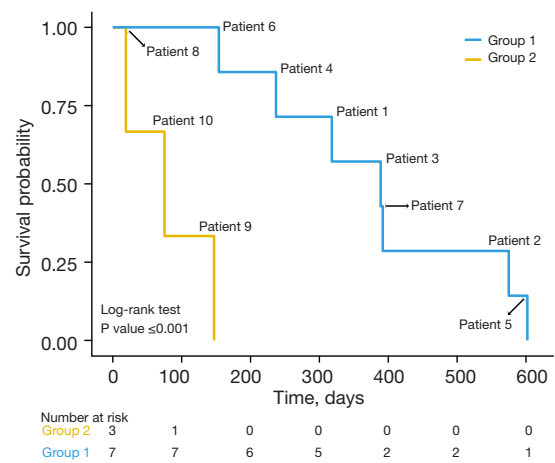


Figure 4 Kaplan-Meier survival plots after the procedure. Group 1: patients treated with rigid bronchoscopy to remove the esophageal stent and implant Y silicone trachea stent; Group 2: patients treated with additional airway stenting without removing the esophageal stent.

hemoptysis, and respiratory insufficiency, while group 2 included severe pneumonia and disease progression. No death was directly attributed to the procedure in both groups.

Discussion

Malignant ERF is a serious and life-threatening event which is common with esophageal cancer and other cancers when invasion of esophagus and/or trachea occurs. Without any therapy, the estimated life expectancy ranges from 1 to 7 weeks (6). These patients usually suffer from repeated aspiration of food, distress due to coughing and shortness of breath, recurrent pneumonia, and poor nutrition, which significantly reduce the quality of life (6). Esophageal stents are commonly used to palliate malignant dysphagia, fistula, or ERF when invasion of esophagus and/or trachea occurs. Prior publications showed that malignant esophageal stent ERF occurs in approximately 9% of patients, which was higher if the patients had undergone radiation therapy (5,13,14). However, little is known about the treatment of this severe complication at present.

The treatment of malignant esophageal stent ERF is challenging. Curative resection of the affected tracheal-bronchial and/or esophageal segments is inappropriate, due to the advanced stage of cancer. Given that most patients are in the terminal stage of their illness, palliative



Video 6 Bronchoscope of new fistula occurrence at the edge of double stent in patient 9.

care becomes the primary treatment objective. Although esophageal bypass procedures can be considered, they are associated with high morbidity and mortality rates and are not suitable as palliative options for advanced cancer (6). The therapeutic goals in these cases are to restore the patency of the trachea, bronchi, and/or esophagus, prevent further material from entering the lungs, and ensure adequate nutrition and fluid intake for the patient.

Currently, endoscopic intervention has been considered as the most effective method for palliation of malignant ERF (6). There are several research reported on the endoscopic treatment of malignant ERF, including airway stents, esophageal stents, double stents, and autologous tissue (15,16). European Society of Gastrointestinal Endoscopy (ESGE) Guideline recommends the application of double stenting (esophagus and airway) when fistula occlusion is not achieved by esophageal or airway prosthesis placement alone (17). Herth *et al.* reported that the survival of airway stents group was significantly lower than the esophageal stents group and double stent group (15). However, whether the treatment for malignant ERF is entirely suitable for malignant esophageal stent ERF remains uncertain (8). Madan *et al.* reported a case of malignant esophageal stent ERF, in which they used a covered tracheal SEMS to seal the ERF through a rigid bronchoscopy (7). This case demonstrated the feasibility of double stenting in the treatment of malignant ERF with indwelling esophageal stent, but the long-term benefit of the patients is unknown.

In this study, we included three double stent patients who placed airway stents without removing the indwelling

esophageal stent. The results showed that this group of patients had lower control of pneumonia and shorter survival compared to those who placed Y silicone stents with esophageal stents removed. The result told us that treatment to malignant ERF and malignant esophageal stent ERF should be different. There may be the following reasons. For patients who develop ERF with indwelling esophageal stents, the esophageal stents were no longer effective tools to seal the fistula, but rather a triggering factor for ERF. If not removed, it will continue to worsen the occurrence of fistulas. Patient 9 is a typical example (Figure S2, Video 6). When we place double stents, we hope that the two stents can precisely seal the position of the fistula while minimizing the antagonistic pressure on the tissue outside the fistula. In this way, the two stents need to be as parallel as possible to reduce angularity. However, this is an idealized idea that is difficult to achieve. Both the esophagus and trachea are active, after long-term swallowing and coughing, the two stents are easily angled, increasing pressure on the tissue of the anterior esophagus and posterior trachea, and leading to tissue necrosis the occurrence of new fistulas (2,9,18). In addition, under the premise that there is already a stent in the esophagus, the placement of the tracheal stent will be limited, and the position of the tracheal stent will be difficult to achieve the expected state, which will affect the sealing effect of the fistula and thus affect the treatment effect. Therefore, we believe that it is necessary to remove the esophageal stent before placing airway stent.

Previous studies on the treatment of malignant esophageal stent ERF have not involved the removal of the indwelling esophageal stent. There may be the following possible reasons for this. On one hand, it is challenging to remove the esophageal stent using traditional gastrointestinal endoscopy or fiberoptic bronchoscopy, as the flexible endoscopes lack sufficient force to accomplish this. On the other hand, there have been no reports on the advantages and disadvantages of removing esophageal stents in these patients. Therefore, many clinicians are hesitant to do so. We are the first to propose the necessity of removing the esophageal stent to show the feasibility and advantage of this procedure.

The removal of esophageal stent and implantation of Y silicone trachea stent were within the same procedure. Silicone stents produce the lowest levels of stress compared with balloon-dilated metal, self-expanding metal, and covered self-expanding metal (19). The weak contact between the silicone stent and the trachea makes them have

higher propensity for migration. In this study, we chose to use Y-shaped silicone stents with the aim of fully leveraging the strengths and avoiding the weaknesses of silicone stents. Choosing a silicone stent instead of a metal stent is to reduce pressure on the edge of the fistula and prevent further expansion. The selection of Y-shaped instead of straight tube silicone stent is to combine with the special anatomical structure of the airway and reduce migration rate. In addition, the radial force of the Y-shaped silicone stent is mainly concentrated in the carina area, which can reduce the dilation force at the edge of the fistula. During the procedure, the edge of the stent extended at least 15 mm beyond the fistula edge. The more extended distance it is allowed, the better. Since the stress of the Y silicone stent is concentrated at the carina and the edge of the stent. Therefore, if the fistula was too large to be covered or the edge of the Y silicone stent can only barely cover the edge of the fistula, we would combine it with a SEMS to extend the edge of the stent, reduce the stress and stimulation on the fistula edge, transferring stress to a distant location.

In this study, we successfully removed the indwelling esophageal stent and implanted a Y silicone trachea stent in all malignant esophageal stent ERF patients. We compare our treatment scheme with double stent, which was recommended in ESGE guideline. The results showed that the patients with esophageal stent removal and Y silicone stent implantation had better sealing effect on the fistula, longer survival, higher quality of life, and better control over pneumonia, compared with patients with double stent. The shortest survival was 154 days in group 1, which is much longer than the reported survival time of malignant ERF (1,2,19). All the patients in group 1 had significant improvement in the degree of dysphagia after the procedure, mainly attributed to the relief of irritating coughing symptoms during swallowing. However, even if the fistula was greatly sealed and the patients have no irritating coughing during swallowing, we recommend them to undergo percutaneous endoscopic gastrostomy or jejunostomy to set up the way of enteral nutrition, to reduce the stimulation of food on the esophageal fistula during repeated swallowing and reduce the risk of further enlargement of the fistula. Four patients in group 1 accepted percutaneous endoscopic gastrostomy or jejunostomy before or after the procedure. All seven patients didn't have irritating coughing during the follow-up time.

There are potential shortcomings of this study. First, due to the nature of retrospective research, some demographic characteristic data were lack, such as the Tumor-Node-

Metastasis (TNM) stage, pathologic pattern of the primary tumor, and imaging materials. Because many patients were diagnosed outside of our hospital, we were unable to obtain these data. During the follow-up process, they expressed uncertainty regarding to this information. Second, the sample size in this study is relatively small. Therefore, further investigations with a larger sample size are warranted to assess the efficacy and safety of this technique. Third, all patients included were male. Since the incidence of esophageal cancer, lung cancer, and gastric cancer were higher among male than female. Further study with female patients of malignant esophageal stent ERF is required.

Conclusions

Removing the esophageal stent and implanting Y silicone trachea stent through a rigid bronchoscopy is a safe and feasible treatment for malignant esophageal stent ERF. This procedure can effectively seal the fistula, prevent from recurrent aspiration pneumonia, improve the quality of life, and prolong the survival time.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://jtd.amegroups.com/article/view/10.21037/jtd-23-1298/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all

aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Ethics Committee of the First Affiliated Hospital of Guangzhou Medical University (approval No. 202050) and individual consent for this retrospective analysis was waived.

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References

1. Shin JH, Song HY, Ko GY, et al. Esophagorespiratory fistula: long-term results of palliative treatment with covered expandable metallic stents in 61 patients. *Radiology* 2004;232:252-9.
2. Bick BL, Song LM, Buttar NS, et al. Stent-associated esophagorespiratory fistulas: incidence and risk factors. *Gastrointest Endosc* 2013;77:181-9.
3. Lenz CJ, Bick BL, Katzka D, et al. Esophagorespiratory Fistulas: Survival and Outcomes of Treatment. *J Clin Gastroenterol* 2018;52:131-6.
4. Grass JK, Küsters N, von Döhren FL, et al. Management of Esophageal Cancer-Associated Respiratory-Digestive Tract Fistulas. *Cancers (Basel)* 2022;14:1220.
5. Park JY, Shin JH, Song HY, et al. Airway complications after covered stent placement for malignant esophageal stricture: special reference to radiation therapy. *AJR Am J Roentgenol* 2012;198:453-9.
6. Kvale PA, Simoff M, Prakash UB, et al. Lung cancer. Palliative care. *Chest* 2003;123:284S-311S.
7. Madan K, Venuthurimilli A, Mohan A, et al. Oesophageal stent-associated esophagorespiratory fistula. *BMJ Case Rep* 2015;2015:bcr2015211880.
8. Rajjman I, Siddique I, Ajani J, et al. Palliation of malignant dysphagia and fistulae with coated expandable metal stents: experience with 101 patients. *Gastrointest Endosc* 1998;48:172-9.
9. Silon B, Siddiqui AA, Taylor LJ, et al. Endoscopic Management of Esophagorespiratory Fistulas: A Multicenter Retrospective Study of Techniques and Outcomes. *Dig Dis Sci* 2017;62:424-31.
10. World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects. *JAMA* 2013;310:2191-4.
11. Mor V, Laliberte L, Morris JN, et al. The Karnofsky Performance Status Scale. An examination of its reliability and validity in a research setting. *Cancer* 1984;53:2002-7.
12. Ogilvie AL, Dronfield MW, Ferguson R, et al. Palliative intubation of oesophagogastric neoplasms at fiberoptic endoscopy. *Gut* 1982;23:1060-7.
13. Parker RK, White RE, Topazian M, et al. Stents for proximal esophageal cancer: a case-control study. *Gastrointest Endosc* 2011;73:1098-105.
14. Homann N, Noftz MR, Klingenberg-Noftz RD, et al. Delayed complications after placement of self-expanding stents in malignant esophageal obstruction: treatment strategies and survival rate. *Dig Dis Sci* 2008;53:334-40.
15. Herth FJ, Peter S, Baty F, et al. Combined airway and oesophageal stenting in malignant airway-oesophageal fistulas: a prospective study. *Eur Respir J* 2010;36:1370-4.
16. Mattioli F, Serafini E, Andreani A, et al. Case report: Endoscopic closure with double stenting and autologous fascia lata graft of large tracheo-esophageal fistula. *Front Surg* 2023;10:1107461.
17. Spaander MCW, van der Bogt RD, Baron TH, et al. Esophageal stenting for benign and malignant disease: European Society of Gastrointestinal Endoscopy (ESGE) Guideline - Update 2021. *Endoscopy* 2021;53:751-62.
18. Nomori H, Horio H, Imazu Y, et al. Double stenting for esophageal and tracheobronchial stenoses. *Ann Thorac Surg* 2000;70:1803-7.
19. Ratnovsky A, Regev N, Wald S, et al. Mechanical properties of different airway stents. *Med Eng Phys* 2015;37:408-15.

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