Clinical Report

Two-tube method for treatment of spontaneous esophageal rupture and concomitant mediastinal infection

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Abstract

Objective: Spontaneous esophageal rupture (SER) is a rare but life-threatening condition with high mortality. The prognosis of patients with SER treated with surgical intervention or the traditional “three-tube” method is controversial. Thus, the aim of this study was to evaluate the clinical efficacy, feasibility, and safety of a new “two-tube” method involving a trans-fistula drainage tube and a three-lumen jejunal feeding tube for the treatment of SER without concomitant pleural rupture.

Methods: From January 2007 to June 2016, patients with SER and managed with the “two-tube” method or other methods were retrospectively analyzed. Data collected included initial presentation, procedure time, duration of treatment, numbers of patients with eventual healing of leaks, and complications.

Results: The average procedure time for the “two-tube” method was 22.1 ± 5.5 minutes. In comparison with the control method, the “two-tube” method had a similar diagnosis time (3.6 ± 1.4 vs. 3.4 ± 1.4 days) but a significantly higher successful closure rate (94.4% vs. 63.6%) and shorter treatment time (38.2 ± 5.6 vs. 53.6 ± 16.9 days). No complications associated with performance of the “two-tube” method occurred.

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Conclusion: The “two-tube” method is an effective and safe approach for patients with SER.

Keywords
Two-tube method, spontaneous esophageal rupture, endoscopic therapy, jejunal feeding tube, minimally invasive treatment, closure rate

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Introduction
Spontaneous esophageal rupture (SER) is a rare but catastrophic condition with high mortality. Current treatment options include conservative treatment, surgical intervention, endoscopic stent placement, thoracic closed drainage, and the traditional “three-tube” method (jejunal feeding tube, gastrointestinal decompression tube, and thoracic closed drainage tube). However, the most effective and feasible treatment option for SER remains controversial, and no standardized treatment algorithm has been established. The present study was performed to introduce and evaluate the clinical efficacy, feasibility, and safety of a new “two-tube” method (trans-fistula drainage tube and three-lumen jejunal feeding tube) for the treatment of SER.

Materials and methods

Study patients
From January 2007 to June 2016, the clinical data of patients who were admitted to our hospital and with a diagnosis of SER were retrospectively analyzed. The diagnosis of SER was made according to one or more of the following criteria: clinical judgment, iodine contrast upper gastrointestinal radiography findings, chest computed tomography (CT) findings, or endoscopy findings. Patients with an esophageal fistula due to postoperative anastomotic leakage or necrosis and those with SER with concurrent pleural rupture were excluded. The ethics committee of Sir Run Run Shaw Hospital approved the study (20171129-1) and waived the need for consent because of the retrospective observational nature of the study.

Equipment
A GIF-Q260J endoscopy system and FG-32L-1 endoscopic foreign body forceps (Olympus, Tokyo Japan), a Freka three-lumen jejunal feeding tube and Freka nasogastric tube (Fresenius Kabi AG, Bad Homburg vor der Höhe, Germany), and a 1000-mA digital subtraction angiography system (Siemens AG, Berlin/Munich, Germany) were used in this study.

“Two-tube” method
Upon confirmation of the diagnosis of SER, the location, size, and number of leaks were further evaluated by CT or iodine contrast upper gastrointestinal radiography. All patients were admitted to the intensive care unit and underwent basic treatments including optimization of perfusion and respiratory function, broad-spectrum intravenous antibiotic therapy, nutritional support, or chest tube drainage. This was usually defined as the traditional “three-tube” method (jejunal feeding tube,
gastrointestinal decompression tube, and thoracic closed drainage tube).

The “two-tube” procedure was accomplished under endoscopy. After the patients had been sedated with intravenous injection of midazolam or propofol, the following procedures were performed. (1) The endoscope was inserted to confirm the location of the leakage and estimate the number and size of the leaks. (2) If the endoscope could enter the leakage cavity, the necrosis within the cavity was directly removed from the cavity. A trans-fistula nasogastric tube was then placed to drain the leakage cavity using an endoscopic foreign body forceps and endoscopic visualization (Figure 1). Otherwise, for patients with a small leak orifice, the drainage tube was placed under X-ray fluoroscopic guidance. The guide wire was first placed in the leakage cavity under X-ray fluoroscopic and endoscopic visualization, and the nasogastric tube was then guided into the leakage cavity. The trans-fistula nasogastric tube was used as a drainage tube for the necrosis in the leakage cavity and was connected to an intermittent continuous vacuum pump at a negative pressure of $-40$ to $-60$ cm H$_2$O. The leak cavity was flushed with normal saline at least twice a day. (3) The three-lumen jejunal feeding tube is a commercially available three-lumen tube. The lumen connected to

Figure 1. Placement of drainage tube into the leakage cavity. (a) Endoscopic view of the leak orifice. (b) Inside view of the cavity under endoscopy. (c) Placement of the drainage tube into the leakage cavity using endoscopic foreign body forceps and endoscopic visualization. (d) Simultaneous placement of the three-lumen jejunal feeding tube.
the most distal tip was used for jejunal feeding, while the other two were used as gastrointestinal decompression tubes (Figure 2). The gastrointestinal decompression tube lumen of the three-lumen jejunal feeding tube was also connected to an intermittent continuous vacuum pump at a negative pressure of $-40$ to $-60$ cm H$_2$O to eliminate gastric fluid, saliva, and phlegm. All patients were given enteral nutrition through the jejunal feeding tube. (4) If necessary, contrast radiography or endoscopy was performed to evaluate the location of the drainage tube and the size of the leakage cavity (Figure 3). The drainage tube was gradually withdrawn according to the amount of drained necrotic tissue, drainage tube location, and the size of the leakage cavity on serial endoscopic or CT check-ups. Usually, 2 to 3 cm of the tube was withdrawn per week. Eventually, when the leakage cavity had closed as shown by contrast radiography or endoscopic examination, the drainage tube was removed from the leakage cavity. Approximately 4 to 6 weeks later, the leakage was usually cured and the leak orifice was closed. A further chest CT scan was performed in necessary to confirm the leakage cavity closure. Finally, the three-lumen jejunal feeding tube was removed when the patient successfully restarted oral feeding. The two indications for surgical management were the presence of sepsis with failure of at least one organ and inefficient drainage through a radiological or endoscopic approach.

**Statistical analysis**

Descriptive data are reported as either mean ± standard deviation, median (interquartile range), or number and percentage. With respect to the differences in outcomes between the “two-tube” method and other management techniques, categorical variables were compared using chi-square analysis. Continuous variables were compared using an independent-sample $t$ test for normally distributed data. A statistical analysis was performed using SPSS 16.0 (Chicago, IL, USA). Significance was defined as a $P$ value of $<0.05$.

**Results**

The “two-tube” method was performed for 18 patients, while 11 patients with SER were managed with control methods. The patients treated with the “two-tube” method comprised 16 men and 2 women. Their mean age was 46.2 years (range, 26–66 years). There were no significant differences in the initial symptoms or underlying comorbidities between the two groups (Table 1). Although the “two-tube” method had a diagnosis time similar to...
that of the control method (3.6 ± 1.4 vs. 3.4 ± 1.4 days), it rendered a significantly higher successful closure rate (94.4% vs. 63.6%, P = 0.033) and shorter treatment time (38.2 ± 5.6 vs. 53.6 ± 16.9 days, P = 0.014) (Table 1).

Two tubes were successfully placed in all cases. The procedure time varied from 15 to 30 minutes (mean, 22.1 ± 5.5 minutes). The treatment cost included the three-lumen jejunal feeding tube, a nasogastric tube, endoscopic examination, and some sedation (total of approximately 250 US dollars). One patient died of severe sepsis and respiratory failure. No complications such as massive hemorrhage or secondary esophageal stricture occurred.

**Discussion**

SER is a rare condition associated with high mortality. The incidence is approximately 0.02%, but the mortality rate ranges from 10% to as high as 60%.³⁻⁵,⁸ It is a life-threatening condition characterized by disruption of the distal esophagus due to barotrauma, resulting in contamination of the mediastinum and pleural cavity with gastric contents. Therefore, early recognition and prompt drainage of
the leakage is of paramount importance. In this study, the “two-tube” method had a significantly higher successful closure rate (94.4% vs. 63.6%, $P = 0.033$) and shorter treatment time (38.2 $/C6$ 5.6 vs. 53.6 $/C6$ 16.9 days, $P = 0.014$) than the control method.

Table 1. Baseline characteristics of patients who underwent the “two-tube” method and other interventions

| Variables                        | “Two-tube” method group (n = 18) | Control group (n = 11) | P value |
|----------------------------------|----------------------------------|------------------------|---------|
| Age (y)                          | 42.6 ± 12.5                      | 54.8 ± 16.6            | 0.127   |
| Male sex                         | 16 (88.9)                        | 9 (81.8)               | 0.592   |
| Initial symptoms                 |                                  |                        |         |
| Vomiting                         | 16                               | 11                     | 0.964   |
| Fever                            | 17                               | 9                      |         |
| Chest pain                       | 18                               | 11                     |         |
| Subcutaneous emphysema           | 10                               | 7                      |         |
| Symptom onset to diagnosis (days)| 3.6 ± 1.4                        | 3.4 ± 1.4              | 0.054   |
| Duration of treatment (days)     | 38.2 ± 5.6                       | 53.6 ± 16.9            | 0.014   |
| Successful closure               | 17 (94.4)                        | 7 (63.6)               | 0.033   |
| Comorbidity                      |                                  |                        |         |
| Coronary heart disease           | 3                                | 3                      | 0.964   |
| Chronic obstructive pulmonary disease | 3                         | 3                      |         |
| Diabetes mellitus                | 4                                | 3                      |         |
| Hypertension                     | 6                                | 4                      |         |

Data are presented as mean ± standard deviation, n (%), or n.

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SER was first described by Herman Boerhaave in 1724; thus, is known as Boerhaave syndrome.4 Because the expected triad of vomiting, chest pain, and subcutaneous emphysema is encountered in only some patients, early diagnosis of SER remains a clinical challenge even now. Patients with SER usually present with atypical symptoms such as respiratory distress, and the physical examination findings are often nonspecific.9,10 Consequently, this entity is often misdiagnosed as a perforated peptic ulcer, acute pancreatitis, acute cholecystitis, myocardial infarction, pulmonary embolism, or spontaneous pneumothorax, which can lead to a delay in the administration of appropriate treatment.8,11 Thus, an accurate diagnosis of SER requires a high index of suspicion, especially in patients with one or two symptoms of the triad.

The treatment of SER depends on the location, size, and duration of the perforation; underlying esophageal disease; and underlying health status of the patient. Adequate drainage and nutritional support are two essential principles in the management of SER.12 Treatment options are categorized into conservative, endoscopic, and surgical. Nevertheless, no consensus regarding the best therapy has been reached. Simple conservative treatments,13 including fasting, intravenous broad-spectrum antibiotics, gastrointestinal decompression, and nutritional support, are generally associated with poor therapeutic effects because digestive fluid may continue to leak into the cavity and the necrotic tissue is not drained. Hence, a thoracic closed drainage tube is placed by most physicians to drain the necrotic tissue and relieve the inflammation. However, given that the leakage and secondary inflammation are mainly located in the mediastinum in most patients,14
placement of a simple closed thoracic drainage tube is not enough to drain the abscessed cavities that are wrapped within the mediastinum. Until 10 years ago, the most commonly used treatment method was a surgical operation, which was thought to thoroughly drain the leakage, pleural cavity, and mediastinal infection. However, such treatment is invasive and associated with high costs and a longer length of hospital stay. Endoscopic clipping is a promising technique for early and small esophageal ruptures. One limitation is that these clips do not allow for treatment of large defects and are not resistant enough to bring together distant, inflamed anastomotic edges. Thus, the efficacy of these techniques is limited by the defect size (generally <2 cm for endoscopic clips). The advent of the endoscopic stent technique revolutionized the management of SER, especially for patients with a small leakage and those unsuitable for open surgery. The mediastinal inflammation may persist, causing the temporary closed leakage to open. Other adverse events associated with stent placement and removal include insufficient closure of the leaks, stent migration and development of strictures after stent removal, uncontrollable hematemesis, and difficult stent removal. Currently, the main therapy for SER is the “three-tube” method, which includes a nasogastric decompression tube, a nasojejunal enteral nutrition tube, and a chest drainage tube. However, the inflammation associated with SER persists for a long time because this management technique does not ensure effective drainage of the mediastinal inflammation and therefore renders a longer length of hospital stay.

We introduced a less invasive but effective “two-tube” method for the treatment of SER. This technique involves the use of a trans-fistula drainage tube and a three-lumen jejunal feeding tube. In our pilot study of 18 patients, 17 patients recovered smoothly and the leakages closed much more quickly than in traditional therapy. The “two-tube” method may have the following four advantages. (1) First, irrigation and drainage can be performed simultaneously. The irrigation frequency and the amount and composition of the irrigation solution can be adjusted according to the characteristics and quantity of the daily drainage fluid. (2) The continuous negative drainage of the tube placed within the leakage cavity may precipitate adherence of the bilateral walls to each other, accelerating closure of the leak cavity. (3) The jejunal nutrition tube enables enteral nutritional support, which not only provides ample nutrition to facilitate closure of the leakage but also decreases the incidence of late inflammation due to bacterial translocation associated with a long fasting period. (4) Finally, thoracic closed drainage is not needed for the vast majority of patients who undergo treatment with our new approach, greatly reducing pain and facilitating earlier mobilization and rehabilitation. Considering the one patient who died of severe sepsis and respiratory failure in the “two-tube” method group, it may be beneficial to continuously lavage and simultaneously drain the abscess cavity with negative pressure. Moreover, for patients with new-onset sepsis or uncontrolled inflammation, an earlier switch to a surgical operation may be an alternative therapy.

In conclusion, the “two-tube” method is an effective, minimally invasive approach to the treatment of SER. The method could be utilized as an alternative therapeutic option for SER.

Author contributions
Liang-liang Yu: Study design, data acquisition, chart abstraction, data analysis/interpretation, manuscript drafting
Zheng-fu He: Chart abstraction, data analysis/interpretation, manuscript revision
Qi-fang Liu: Data analysis/interpretation, manuscript revision
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Jian-cang Zhou: Study concept and design, data analysis/interpretation, manuscript revision

Declaration of conflicting interest
The authors declare the there is no conflict of interest.

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