

Brief Technique Reports

Surgical Management Of 3 Cases With Huge Tracheoesophageal Fistula With Esophagus Segment *in situ* As Replacement Of The Posterior Membranous Wall Of The Trachea

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ABSTRACT **Objective:** Tracheoesophageal fistula (TEF) is an uncommon and potentially life-threatening complication of blunt chest trauma. The objectives of this report are to describe our surgical experience in three patients with huge TEF and to evaluate the short-term results of surgical management in this potentially life-threatening complication of blunt chest trauma.

Methods: Three patients with huge TEF (5.2-7.0 cm in diameter) after blunt chest trauma were kept in supine position, then the neck was excised and esophagus was free, double breasted suture and clench and mutilation were performed successively over the same level of superior aperture of thorax. The thoracic esophagus was then located *in situ* at the membrane portion where the trachea was incomplete. Meanwhile, the stomach was then freed through a middle abdominal incision and pulled through the posterior tunnel of the sternum to the neck. Last, an anastomosis between esophagus and tubular stomach was performed over left neck.

Results: All three patients recovered well after the operation. They were able to take liquids and then solids beginning ten days after the procedure. One year post-operation, they were able to resume normal activity.

Conclusions: The surgical management of patients with huge TEF by esophageal exclusion (cervical gastroesophagostomy) and use of esophagus segment *in situ* as replacement of the posterior membranous wall of the trachea is feasible.

KeyWords: huge tracheoesophageal fistula; Surgical management

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Introduction

Tracheoesophageal fistula (TEF) is a very uncommon and potentially life-threatening complication of blunt chest trauma. The first reported case of TEF caused by blunt chest injury was published in 1936 by Vinson (1). Beal et al. have estimated the incidence of blunt esophageal perforation in major trauma to be as low as 0.001% (double check, if is true, then delete this sentence since the morbidity is too low) (2). Clinically, coughing and choking after swallowing, the so-called "swallow-cough complex" or Ono's sign, is considered a classic sign for the presence of TEF. Confirmation and extent of TEF can be accurately diagnosed on esophago-

graphy and CT imaging (3,4), while supplemental information may be acquired with esophagoscopy and bronchoscopy (5).

After diagnosis is confirmed, surgical repair must be carried out as soon as possible. Non-operative mortality is as high as 80% while operative mortality is estimated at 9.3% (6). Closure of the membranous trachea in one layer and closure of the oesophagus in two layers, which are membrane and muscle layer, is commonly undertaken (7). Structural interposition between the trachea and oesophagus is advised to decrease the recurrence of TEF and can be achieved either with a muscle (intercostals, sternothyroid, sternocleidomastoid, strap) or pericardial/pleural flap to separate the lumen of trachea and esophagus (6,8-12) as well as with fibrin glue being applied together (13). However for patients with a huge TEF, that is TEF greater than 5 cm, surgical repair is difficult to perform as the huge defect and necrosis of local tissues after the blunt chest trauma may be accelerated with concurrent infection and chemical erosion.

While esophageal excision is useful in patients with simple esophagus fistulas the reconstruction of the trachea will be more difficult and the risk of failure in patients with TEF is increased due to vascular damage to the tracheal tissue. Further, the use of esophageal excision with anastomosis between tubular stomach

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and the esophagus above the fistula, reported by Steiger et al., is well described as a classical palliative surgery in patients with malignant TEF (14). Here we describe the successful treatment of three patients with huge TEF after blunt chest trauma by esophageal exclusion (cervical gastroesophagostomy) and use of esophagus segment in situ as replacement of the posterior membranous wall of the trachea. The objectives of this report are to describe our surgical experience in three patients with huge TEF and to evaluate the short-term results of surgical management in this potentially life-threatening complication of blunt chest trauma.

Patients and methods

We studied three patients with huge TEF after blunt chest trauma who were successfully treated by esophageal exclusion (cervical gastroesophagostomy) and use of esophagus segment in situ as replacement of the posterior membranous wall of the trachea.

Case 1.

A 21-year-old male was referred to our department in February 2007 from a hospital in Macao, PR China, due to multiple traumas sustained in an automobile accident thirteen days prior.

Patient was initially seen for extensive traumatic injuries in the Emergency Department at a local hospital in Macao. Internal fixation of bilateral lower extremities fracture as well suture and bandage to numerous wounds were performed. After his surgical procedures, his extremities were noted to move freely but he then developed shortness of breath, hypotension, and vascular insufficiency. He then was intubated and placed on mechanical ventilation to sustain his vital functions. Radiographic and clinical analysis demonstrated bilateral tension pneumothorax, hemothorax and mediastinal emphysema. After performing bilateral thoracic and mediastinal cavity drainage the circulatory system became stable but his ventilation remained unstable. Further investigation with bronchoscopy and CT imaging showed an approximate 1cm TEF as well as extensive chemical burn of lungs caused by erosive gastric reflux (Fig 1A and 1B). A subsequent jejunostomy and gastrostomy was performed to provide enteral alimentation as well as decompression of the stomach. However, erosive reflux continued and the patient developed a secondary pulmonary infection due to aspiration of gastric contents. Further, as the initial TEF was proximal to the carina the patient continued to suffer unstable ventilation. Ultimately, a left double-lumen tracheal catheter was inserted into left bronchus to obtain left-lung unilateral pulmonary ventilation (Fig 1C). There after the patient was referred to our facility for further evaluation and treatment.

Initial evaluation by bronchoscopy and esophagoscopy in our department demonstrated a huge TEF. A biliary fistula was confirmed with erosion of the trachea and tunica muscularis identified. The size of fistulae enlarged from 1cm to 7cm over 72 hours. Complete loss of the posterior wall of thoracic segments of pars mem-

branacea of trachea and esophagus was noted along with the plane encompassing the superior aperture of thorax to 1 cm above carina was absent and the region fused into a huge fistulae. The case was further complicated at this point with extensive bilateral pneumonia with urulent sputum noted in the trachea (Fig 1C). A left-lung unilateral respirator with left branch of double tubes bronchial catheter was maintained for ventilation. Now a common cavity between the esophagus and trachea measuring 7cm in length was detected through thoracic CT scans (Fig 1D).

After sufficient preoperative preparations, surgical repair was performed under general anesthesia. Surgical exploration indicated "frozen mediastinum" due to inflammation and the plane from the superior aperture of thorax to the carina could not be separated. Extensive collateral vascularization was noted over the esophagus and trachea all associated with superior mediastinal inflammation. We noted the dislocated esophagus for blood supply over internal segment posterior wall of thorax. We then took the dislocated esophagus over internal segment posterior wall of thorax and replaced with the membrane of trachea. The esophagus was severed from 2 cm below the carina, then using double breasted suture and clench, the thoracic cavity was closed.

The patient was kept in supine position, then the neck was excised and esophagus was free, double breasted suture and clench and mutilation were performed successively over the same level of superior aperture of thorax (Ethicon, Johnson & Johnson, Pennsylvania, USA). The thoracic esophagus was then located in situ at the membrane portion where the trachea was incomplete. Meanwhile, the stomach was then freed through a middle abdominal incision and pulled through the posterior tunnel of the sternum to the neck. Last, an anastomosis between esophagus and tubular stomach was performed over left neck.

Case 2.

A 6-year-old boy was referred to our department from a hospital in Shanghai, PR China, due to choking after taking food. Six months prior to referral the child was severely injured in an automobile accident and during recovery he required a tracheotomy and gastrostomy for five and a half months. He was initially evaluated and diagnosed with a traumatic TEF by esophagoscopy and bronchoscopy in an Emergency Department Anhui province, PR China. He was then referred to a children's hospital in Shanghai, PR China where tracheotomy and gastrostomosis were performed to stabilize his ventilation and nutrition. After stabilizing and receiving symptomatic treatment his condition improved. He was then referred to our department for further evaluation and treatment.

On admission we noted the child to be thin and small, with a weight of 15 kg. On examination, the tracheostomy cannula was unobstructed, the inferior extremity of which reached at the level of carina; on auscultation moist and coarse rales in the lungs were noted; the gastric stoma duct over abdomen was unobstructed, and

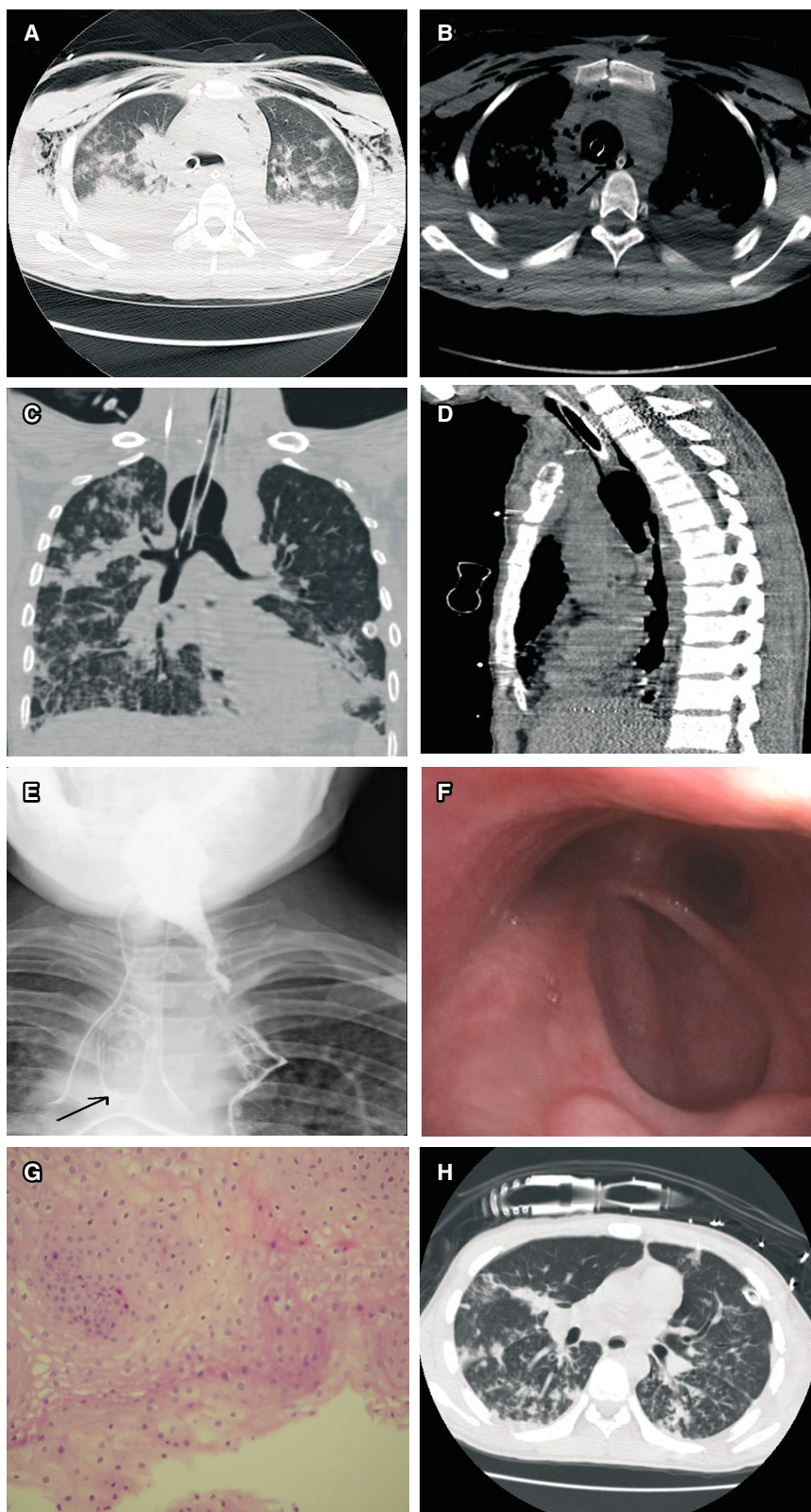


Fig 1. Figures of Case 1. A. Chemical burn and secondary infection in both lungs due to reflux of gastric contents; B. Computed tomogram of the thorax shows the tracheoesophageal fistula only approximately 1.0 cm (arrow); C. The intubation with left double-lumen tracheal catheter into left main bronchus; D. The cavity between esophagus and trachea was 7 cm in diameter; E. A residual cavity remains post operation; F. The residual cavity cured well after three months of operation; G. Only a few inflammatory cells founded in mucosa of the excluded esophagus; H. Lung chemical burns reduced after drainage by gastrostomy compared with the Fig A.

fluid nutritional diets could successfully pass through gastric stomach. After bronchoscopy and three-dimensional reconstruction examination of chest CT scan, a diagnosis of huge TEF was confirmed with the fused common cavity between esophagus and trachea measuring 5.2 cm in length. The child's case was further complicated with bilateral lower lobe pneumonia associated with gastric reflux and aspiration.

Prior to definitive repair a successful jejunostomy was performed to maximize nutrition and prevent further aspiration. Enteral nutrition was administered via a jejunal fistula. The gastric fistula was then connected via an external vacuum extractor to reduce pulmonary inflammation caused by chronic gastric reflux into lungs. The patient's condition significantly improved with supportive care and his weight began to increase. After a month of supportive care and with careful preparation, replacement of the membranous trachea with dislocated esophagus in situ and esophageal-gastric anastomosis over the left neck via a meta-sternal tunnel were performed through the two incisions at neck and upper central abdomen.

Case 3.

A 46-year-old woman was referred to our department on August 20, 2008, from a hospital in Shenzhen, PR China, because of multiple traumas over the whole body and bucking for a month, caused by traffic accident. Four months before, patient was admitted to a hospital in Shenzhen because of multiple traumas caused by traffic accident. She was found to be with bilateral pulmonary contusion, fracture of left ribs from the third to seventh, right ribs from second to eleventh, left collar bone, and transverse process in the right side of C7. Bilateral thoracic close drainage was performed in the emergent salvage. After the operation, she was sent to ICU and given respirator to assist ventilation because of hemorrhagic shock. Further examination CT scan showed right pneumothorax, consolidation of right lower lung and contusion of left lower lung.

Patient's inflammation and pulmonary contusion were improved after anti-inflammatory therapy and other symptomatic treatments. Pleural canals in her thoracic cavities were pulled out 2 weeks later. One month later, the respirator and tracheal intubation were moved away; and patient's condition of spontaneous ventilation was good. But meanwhile, patient presented with edema of right lower extremity. Colour Doppler Ultrasound examination indicated right femoral vein thrombosis; and her symptom was reduced after receiving anticoagulant therapy. Two months later, there was no abnormality of the pulling out of gastrointestinal decompression tube and lipid diet.

Forty days before, patient was moved to ICU because of aspiration pneumonitis caused by reflux of gastric contents, which was improved a week later, and control of pulmonary infection was continued to be performed. Vomiting and bucking re-occurred a month before when he drank water. Posterior wall of the superior segment of trachea and unclear boundary with esophagus were

found through chest CT scan. He was admitted to our hospital for further consultation because he was suspected to be with esophago-tracheal fistula.

When she was admitted: little thin and small figure, smooth respiration about 26/min, bondage of moist and coarse rales could be heard over lungs; abdomen was flat and soft without palpable tumor or tenderness; myodynamia of both upper extremities was about from grade II to III, and that of both lower extremities was from grade IV to V. Nutrient canal of duodenum was unobstructed, and tube feeding with fluid food was normal, but there was lots of white spumy sputum. Esophago-tracheal fistula was accurately diagnosed through bronchoscopy and chest CT scan reconstruction. Esophagus and trachea was contacted for about 5.3cm.

After admission, her nutritional condition and clinical symptoms were improved through overall check up, nutritional support, pressure sore prevention attendance, anti-inflammatory and anticoagulant therapy and other symptomatic treatments. Then, through the two incisions at left neck and upper abdomen, replacement to membranous part of trachea with dislocated esophagus, esophagus and gaster anastomosis over left neck through the meta-sternum tunnel was performed on October 9, 2008 under systemic anesthesia. The detail of this case was previously report (15).

Results

The case 1 recovered well after the operation. The tracheal intubation tube was removed 24 hours after the operation and the patient could expectorate on his own. He was able to take liquids and then solids beginning ten days after the procedure. Broad spectrum antibiotics were given and imaging showed resolution of pneumonia. Esophagogram and tracheography were performed and indicated a persistent cavity connecting with trachea over dislocated oesophagus (Fig 1E). He was discharged home on post-operative day twenty. He was seen in follow-up regularly in our department. The status of ventilation was normal, tracheoscopy indicated trachea was unobstructed, and residual cavity of the dislocated oesophagus was well healed (Fig 1F). Pathology showed only scant inflammatory cell infiltration over mucous membrane of the esophagus six months post operation. One year post-operation, his activity level was normal (Fig 1G). At fifteen months post operation his lung function indicated: FVC77.20%, FEV1 84.58%, FEV1/FVC 92.1; airway resistance guideline total resistance of respiration is 136%, viscosity resistance under 5Hz was 132%, and there was no evidence of gastric reflux into the esophagus using a 24-hour continuous PH monitor at the gastro-esophageal anastomotic stoma.

For the case 2, tracheal intubation was discontinued two hours after the operation and the patient could breathe and expectorate on his own. He was able to take an oral diet by post-operative day 10 and he was discharged home eighteen days later. Bronchoscopy performed four months after the operation indicated mucous membrane of esophagus replacing membranaceous tracheae satisfactorily. The patient was able to resume normal activities includin

g returning to school. Nine months after the operation, his pulmonary function tests indicated: FVC55.00% , FEV1 60.76% , FEV1/FVC 90.52%; airway resistance guideline: total resistance of respiration was 134%, viscosity resistance under 5Hz was 130%. A 24-hours PH monitor evaluation at the gastro-esophageal anastomotic stoma could not be performed as the young patient was unable to cooperate with the examination.

And for the case 3, trachea cannula was removed an hour later after the operation, and patient can breathe and expectorate on her own. Neck incision became red and swollen on the sixth day after the operation, which was improved after change of dressing and drainage. On the 20th day after operation, her taking food became normal and she was discharged 24 days later after operation.

Discussion

TEF complicates the course of disease in about 20% patients with esophageal and tracheal cancer (16). It is estimated that only 10% of acquired TEF are due to benign disease (17). TEF is a rare complication following blunt chest trauma. In a retrospective study of 3,606 trauma patients reported by Beal and associates (2), 2,560 patients (71%) sustained blunt trauma and only 3 patients (0.001%) developed esophageal perforations. One could postulate, therefore, that the incidence of TEF probably constitutes less than 0.001% of all blunt trauma cases. An extensive review of the literature by Reed et al (6), includes 61 cases since the first reports by Vinson in 1936 (1). Accordingly, the commonest mode of injury is vehicular accidents involving an unrestrained driver and the most common site of fistula formation is in the posterior trachea at or just above the carina (6).

The majority (80.4%) of TEFs became symptomatic within the first 10 days of injury with immediate onset in 11.5% of the cases (1). Late and very-late onset of TEF after trauma has also been reported as late as 425 days after injury (18), 20 years (9), or even 38 years (19). Immediate onset is often associated with tracheal and esophageal rupture with subsequent fistula formation. In late onset, the mechanism may be slightly different with compression of the trachea and esophagus between the sternum and the vertebral bodies resulting in partial laceration of the posterior membranous trachea with contusion and loss of vascularity to the adjacent anterior esophageal wall. Subsequently, necrosis of the esophageal wall occurs over time resulting in a delayed TEF (6).

Several mechanisms also have been proposed to explain TEF formation (20-22). In Case 1, we noted the fistula enlarged from 1cm to 7cm within three days. We believe the mechanism of TEF formation in this case may be compression of the trachea and esophagus between the sternum and vertebral bodies. We note the reflex closure of the glottis together with thoracic compression can produce a rapid increase in intraluminal pressure, which would be greater in the larger airways. Subsequently, a partial laceration occurs in the mucosa of the posterior membranous trachea, resulting in reflux bile erosion to the trachea, from the mucosa to submu-

cosa, basement membrane, and muscle of trachea, which can't be protected by the mucosa due to the laceration in the mucosa. Concurrently, the anterior esophageal wall is damaged with impairment of the mucosal blood supply. Esophageal necrosis then occurs rapidly and followed by TEF formation. It is interesting to find the trachea and esophagus merge with single huge cavity. (Fig 1C,D) The delayed formation of the TEF could account for the majority (59.0%) of patients who exhibit symptoms 3 to 10 days after blunt trauma (6,23,24). However, Stothert and associates propose an alternate mechanism of TEF formation involving immediate tracheal and esophageal rupture with subsequent fistula formation (21).

Coughing and choking after swallowing, the "swallow-cough complex" or Ono's sign, is considered the classic sign of TEF. Other signs and symptoms include crepitation over the neck, chest, or abdominal pain, hemoptysis or hematemesis, dyspnea, dysphagia, hoarseness, odynophagia, and abdominal distention. The most commonly associated findings reported include subcutaneous air (54.1%) and pneumothorax or pneumomediastinum (41.0%). Rib fractures (31.1%) and hemoptysis (26.2%) are also commonly reported findings (6), as well as the "breathing-bag sign" (25). Once the diagnosis of TEF is suspected, mediastinal contamination should be minimized by placing a large nasogastric tube in the patient's stomach and another in the proximal esophagus to remove secretions. The presence and extent of the TEF can then be evaluated and diagnosed on esophagography and CT imaging (3,4). Supplemental information may also be acquired with esophagoscopy and bronchoscopy (5). In addition to direct signs of TEF, imaging can identify associated problems such as pneumonia, gaseous distention of the esophagus, pneumomediastinum, and subcutaneous air.

Adequate enteral or parenteral nutrition should be initiated in perioperation period. Fluid and electrolyte imbalances should be corrected if necessary, and patients should receive broad-spectrum antibiotic prophylaxis (6). In both our cases, patients suffered serious consequences due to reflux and aspiration through the TEF. Often a staged operative recovery is necessary. The aim of the first-stage is to exclude oesophageal contents from the respiratory tree. Gastrostomy and negative pressure drainage can reduce dangerous gastric content reflowing and aspiration.

After diagnosis of TEF is confirmed, surgical repair should be carried out as soon as possible. Currently, non-operative mortality is 80% while operative mortality is 9.3% (6). Closure of the membranous trachea in one layer and closure of the oesophagus in two layers should be undertaken (7). Structural interposition between the trachea and oesophagus is advised to decrease the recurrence of TEF. This is achieved either with a muscle (intercostals, sternothyroid, sternocleidomastoid, strap) or with a pericardial/pleural flap to separate the lumen of esophagus and trachea (6,8-12,38-43), or with fibrin glue being applied together (13). The size of TEF is 0.5 to 3.0 cm in length for most patients. However, for patients with so-called huge TEF (> 5 cm in length) such procedures are more difficult due to necrosis of local tissues associated with infection-

ter blunt chest trauma. In these cases, the tension of the muscle or pericardial/pleural flap will not be enough as the posterior membranous wall of the trachea to sustain the trachea to preserve the normal respiration in patients with huge TEF. Esophageal excision is useful in patients with simple esophagus fistula but the reconstruction of trachea is more difficult, the risk of failure in patients with TEF is very high, and because the vascular supply of the trachea is often damaged. Esophageal excision with anastomosis between tubular stomach and the esophagus above the fistula, reported by Steiger et al. is described as classical palliative surgery in patients with malignant TEF (14). A review of the literature, demonstrates only one case of surgical repair involving esophageal resection with colonic interposition in patients with TEF after the blunt chest trauma (44).

In this study, we define huge TEF as TEF greater than 5 cm in length. In another review by Freitag et al., the fistulas from 1 to 4 cm in length were defined as "very large fistulas" (45). We believe that operative decisions regarding management of TEF should consider both the size of fistula as well as the timing of the diagnosis. Early diagnosis is critical to obtaining ideal results and prompt treatment as well as structural interposition between the trachea and oesophagus to decrease the recurrence of TEF. Delayed diagnosis of TEF, especially when the trachea and esophagus merge into a huge cavity as in our two cases presented, make surgical repair even more difficult.

We successfully treated three patients with huge TEF after blunt chest trauma by esophageal exclusion (cervical gastroesophagostomy) and incorporated esophagus segment in situ as replacement of the posterior membranous wall of the trachea. To our knowledge this is the first report of this method to treat patients with huge TEF after blunt chest trauma. The risk of operation was decreased greatly since this direct and simple procedure not only blocked the reflux to trachea, also avoided the further surgical intervention in the inflammation, edema and necrosis area. Furthermore, the trachea and the tube stomach were separated after reconstruction since the tube stomach was located in anterior mediastinum, but trachea in postmediastinum, this was also avoid the change of TEF recurrence. By using this surgical method to treat the huge TEF, two questions have to be considered. Firstly, whether the remained esophageal cavity in situ affect the patient's pulmonary function short term and long term. Secondary, whether the secretion from remained esophageal cavity in situ continually cause the pulmonary infection. In our three cases, self-breathing was achieved at 1, 2 and 24 hours after removing tracheal intubation, respectively. In case 1, the chest tube was removed 72 hours after the operations. Patients began oral nutrition 9 days, 12 days and 20 days after the operation and were discharged home on day 18, 22 and 24 after operation, respectively. These cases have been followed now for 2, 18 and 23 months, respectively, and all patients have resumed normal activities of daily living. Pathology indicated mucous membrane of esophagus was with good activity, there was only a little inflammatory cell infiltration. The lung function and air-

way resistance recovered well during the follow-up on 15 and 9 months later in case 1 and 2, respectively. In case 1, no sign of gastric reflux in esophagus was seen using 24-hours PH monitor around gastro-esophageal anastomotic stoma. However, these results need to be further studied and confirmed by long term follow-up. An obvious limitation of our report is that there are only three cases to describe from a single institution and this should be evaluated in more cases studies from multiple centers.

In summary, huge TEF is rare and potentially fatal complication of blunt chest trauma. In our three cases, the surgical management of patients with huge TEF by esophageal exclusion (cervical gastroesophagostomy) and use of esophagus segment in situ as replacement of the posterior membranous wall of the trachea is feasible. Improving the nutrition status and controlling the lung infection were critical in perioperation period. We emphasize that the risk of operation and TEF recurrent were decreased greatly since this direct and simple procedure avoided the further surgical intervention in the inflammation, edema and necrosis area.

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