Surgical management of acquired tracheo/bronchoesophageal fistula associated with esophageal diverticulum

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Background: The reports on acquired tracheoesophageal fistulas (TEFs) or bronchoesophageal fistulas (BEFs) associated with traction esophageal diverticula (TED) are rare. Here, we present our experience of six cases.

Methods: Between Jan. 2015 and Jun. 2016, 6 patients were admitted to our department for TEF/BEFs combined with esophageal diverticula. Clinical data of the 6 patients were retrospectively reviewed.

Results: All orifices of TEF/BEF in the esophagus side opened at the diverticula wall. The orifices in the airway side were 2 at the carina and 4 at the right intermediate bronchus. All six patients received the same intervention: a limited diverticulectomy with the fistula resection was done in the esophagus; separate layers of repair were performed for the defect in the esophagus; the muscle flap interposition was used in all six cases. All postoperative courses were uneventful. No recurrence fistula and symptomatic diverticula occurred. The airway and esophagus were patency during a median of 9-month follow-up.

Conclusions: Acquired TEF/BEFs caused by esophageal diverticula can be treated successfully by surgery. A limited diverticulectomy is sufficient to ensure enough esophagus remodeling.

Keywords: Tracheo/bronchoesophageal fistula (TEF/BEF); esophagus diverticulum; acquired

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Introduction

Acquired tracheoesophageal or bronchoesophageal fistulas (TEFs or BEFs) is a rare disorder with a variety of conditions. The majority causes of acquired non-malignant TEFs or BEFs fistulas include postintubation injury, chronic infections (e.g., tuberculosis), radiation injury and postsurgical lesions (1,2). Traction diverticulum results from external inflammatory reactions in neighboring mediastinal ganglia, the tissue adhesion to the esophagus that contracts and scars the wall of the esophagus to provoke

the diverticulum. In rare cases, this causes inflammatory necrosis, and a fistula will appear between the diverticulum and the respiratory airways (3).

Surgical intervention for TEF/BEFs fistulas caused by diverticulum has been especially rare. From Jan 2015 to Jun 2016, six consecutive cases of acquired TEFs/BEFs through esophagus diverticulum were admitted to our institute and underwent operation. We review the presentation and management of six patients and include a literature review with a summary of surgical indications and operational techniques.

Methods

Approval for the study was obtained from the Ethics Committee of Shanghai Chest Hospital [KS(Y) 1723]. Written informed consent was obtained from each patient or her legal representative.

Case descriptions

Case 1

The 67-year-old female patient developed a persisting cough while eating and signs of aspiration pneumonia for 6 months. The patient had a history of mid-esophageal diverticulum diagnosed for 30 years, with the symptom of regurgitation. An acquired TEF 0.5 cm proximal to the carina was detected on esophagoscopy and bronchoscopy. Upper GI series using gastrografin showed that the diverticula combined with a fistulous tract between the esophagus and the bronchial tree. A chest CT revealed pneumonia in the bilateral lung and bronchiectasis in the right low lobe. The patient received antibiotic treatment before operation.

Case 2

A 47-year-old female was admitted to our center with persistent cough of 2 years. Esophagoscopic evaluation demonstrated an orifice in the esophageal diverticulum, but bronchoscopy displayed normal. The esophagogram showed the leakage of gastrografin, and a chest CT scan revealed an image of contrast medium in the bronchus of the right low lobe right after esophagogram.

Case 3

A 78-year-old female patient was repeatedly hospitalized for persistent cough during the recurrent pulmonary infection over a period of 72 months. Esophagoscopy and bronchoscopy revealed a TEF 0.4 cm proximal to the carina. Esophagogram also showed the tract between the esophagus and airway conduit. CT scan showed severe bilateral pneumonia, and more serious in left inferior lobe. A nasogastric tube was inserted for nutrition intake and antibiotic treatment was managed on account of the pneumonia

Case 4

The 51-year-old female complained with cough after swallowing for 2 months. Esophagoscopy discovered a diverticulum 30 cm away from upper incisor in the anterior wall of esophagus, and an orifice in the top of diverticulum. The esophagogram imaging exhibited contrast medium leakage to intermediate bronchus, and CT examination indicated bronchiectasis in inferior lobe of right lung.

Case 5

The patient is a 68-year-old woman with diverticulum who complaint of a racking cough while eating and following chest tightness for 36 months. A fistula between esophagus and right intermediated bronchus was showed in upper GI series; esophagoscopy detected a diverticulum 25 cm away from upper incisor and an orifice in the top of diverticulum. Bronchoscopy evaluation showed obvious mucosa hypertrophy of intermediate bronchus and atelectasis of inferior and middle lobe of right lung was confirmed by chest CT scan.

Case 6

A 68-year-old female patient was confirmed BEF combined with diverticulum for 24 months and failed with two-times esophagus stent insertion and one time clip using. Nutrition intake was through a nasogastric tube for 12 months before admitting in our division. Orifice of fistula in diverticulum was 25 cm away from upper incisor through esophagoscopy evaluation. Esophagogram also showed the contrast medium tract, and CT scan showed pneumonia in inferior lobe of right lung.

Table 1 showed all patients' pre-operative demographics, and *Figures 1-6* showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram for patients 1–6.

Surgical manipulations

Dissection of the diverticulum and the fistulous tract was conducted through the right thoracotomy. Limited diverticulectomy and the adjacent inflammatory mass resection were performed. The fistulas were removed with suture closure of the tracheal/bronchus defects or lobectomy. Esophageal mucosa and muscle were separated and sutured for esophagomyotomy. Vascularized muscle flaps were interposed between the repaired esophageal and tracheal/bronchus defects.

Postoperative complications and mortality

Adverse events occurring in hospital within 30 days after surgery were defined as postoperative complications. Operative mortality was defined as death occurring within 30 days of the operation.

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Table 1 Patients	' characteristics
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Characteristics	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Age	65	46	78	51	68	54
Gender	Female	Female	Female	Female	Female	Female
Symptom	Cough during eating	Persist coughing	Coughing with swallowing	Cough during eating	Cough and oppression in the chest	Cough during eating
Duration of symptom (months)	6	24	72	2	36	60
Physical examination	Bilateral pulmonary rale	Lung rale in RLL	Bilateral pulmonary rale	Lung rale in RLL	Lung rale in RLL	Normal
Esophagoscopy						
Location of diverticulum (away from upper incisor, cm)	26	30	28	30	25	23
Orifice of fistula (away from upper incisor, cm)	28	31	29	32	26	25
Bronchoscopy	Orifice in trachea 0.5 cm proximal to the carina	Normal	Orifice in trachea 0.4 cm proximal to the carina	Normal	Mucosa hypertrophy of intermediate bronchus	Normal
Upper GI series by using gastrografin	Leakage to bilateral pulmonary	Leakage	Leakage to RLL	Leakage	Leakage	Leakage
Chest CT scan	Pneumonia	Contrast medium in right intermediate bronchus	Pneumonia	Bronchiectasis in RLL	Atelectasis in RIL + RML	Pneumonia in RIL
FEV1 (L)	1.86	2.11	1.78	1.83	1.29	2.17
FEV1/FVC	89%	96.3%	91%	75.2%	59.9%	92.9%
DLCO-SB	59.7%	73.1%	63.2%	95.1%	63.9%	113.4%

RIL, right low lobe; RML, right middle lobe; FEV1, forced expiratory volume in one second; FVC, forced vital capacity; DLCO, carbon monoxide diffusing capacity.

Results

Surgical procedure and postoperative outcomes

Six patients received surgical therapy after improvements in nutritional status. During the operation, the surgeons found that the orifice airway conduit was located in the tracheal approach to the carina for two patients and in the intermediate bronchus for four patients. The orifice of the esophagus conduit was located in the diverticula for all patients. Limited diverticulectomy and the tracheal/ bronchus defects were repaired in five patients, and limited diverticulectomy with lobectomy of right lower lobe (RIL) + right middle lobe (RML) were performed in case 5, causing of the obvious mucosa hypertrophy of intermediate bronchus and completely atelectasis in RIL + RML. Intercostal muscles flap in two patients and serratus anterior muscular flaps in four patients were used to separate the esophagus and trachea/bronchus.

Operative bleeding ranged from 90 to 220 mL, and operative time ranged from 95 to 210 min. Pathologic examination of the resected specimen showed chronic inflammatory tissue surrounding the resected esophageal diverticulum and the adjacent bronchus. A nasogastric tube was placed at the duodenum for enteral nutrition. The esophagography was successfully performed on the 7th postoperative day for all patients and no leakage from the esophagus was noted. Then, patients were encouraged to sip water and the oral intake were gradually increased until the patient could tolerate soft diet without parenteral nutrition. The length of hospital stay ranged from 8 to 20 days (post-operative information was showed in Table 2, No. (D) image of Figures 1-6 showed the post-operative esophagogram). No postoperative complication or mortality was observed.



Figure 1 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 1. The arrows in the No. (A-C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view, the orifice in airway lumen through bronchoscopic view and the leakage of contrast medium respectively.



Figure 2 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 2. The arrows in the No. (A,C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view and the leakage of contrast medium respectively. No. (B) image showed normal bronchoscopy.

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Figure 3 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 3. The arrows in the No. (A-C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view, the orifice in airway lumen through bronchoscopic view and the leakage of contrast medium respectively.



Figure 4 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 4. The arrows in the No. (A,C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view and the leakage of contrast medium respectively. No. (B) image showed normal bronchoscopy.



Figure 5 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 5. The arrows in the No. (A,C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view and the leakage of contrast medium respectively. No. (B) image showed mucosa hypertrophy of intermediate bronchus.



Figure 6 The images (A-D) showed the esophagoscopy, bronchoscopy, pre- and post-esophagogram of patient 6. The arrows in the No. (A,C) images displayed the orifice in esophageal diverticulum under the esophagoscopic view and the leakage of contrast medium respectively. No. (B) image showed normal bronchoscopy.

Table	2	Post-o	peration	information

Variables	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Insider diameter of fistula (mm)	1.5	2	2	1.5	2	1.5
Length of fistula (mm)	5	6	4	5	5	6
Location of fistula in air conduit	Trachea	Intermediate bronchus	Trachea	Intermediate bronchus	Intermediate bronchus	Intermediate bronchus
Surgical approach of trachea/bronchus defective	Limited diverticulectomy and closure trachea defects	Limited diverticulectomy and closure bronchus defects	Limited diverticulectomy and closure trachea defects	Limited diverticulectomy and closure trachea defects	Limited diverticulectomy and Lobectomy of RML + RIL	Limited diverticulectomy and closure trachea defects
Surgical approach of esophagus defective	2-layer closure	2-layer closure	2-layer closure	2-layer closure	2-layer closure	2-layer closure
Muscle flap interposition	Serratus anterior	Intercostal muscles	Serratus anterior	Intercostal muscles		Serratus anterior
Operation time (min)	180	210	155	150	160	95
Blood loss (mL)	150	175	220	100	150	90
ICU stay (days)	1	1	3	1	1	2
Chest tube duration (days)	8	8	12	4	6	7
Transfusion (U)	0	0	0	0	0	0
Hospitalization (post- operation)	12	8	20	10	13	8
Complications	No	No	No	No	No	No
Date of operation	Jan. 28, 2015	Oct 16, 2015	Jan 1, 2016	May 9, 2016	May 20, 2016	Jun 6, 2016
Follow-ups (months)	21	12	9	5	5	4

Follow-up information

Complete follow-up information was obtained for six cases. During a median follow up of 9 months, no patient experienced symptoms of fistula reoccurrence or esophageal stenosis.

Discussion

Benign BEFs in adults are uncommon; the most frequent etiologies for the fistulae are complications of postintubation injury and prior esophageal surgery. Traction esophageal diverticula (TED) is one kind of true diverticula that develops as the results of chronic inflammatory contracture from mediastinal structures outside the esophagus. Most TEDs occur in the upper middle of the thoracic esophagus in a peritracheal/bronchial location (4). Diverticula of the esophageal body comprise 10% to 15% of all esophageal diverticula. The natural history of midesophageal diverticula may lead to severe complications, such as erosions of the esophageal wall, and cause hemorrhage or perforation (5). Surgical approach for acquired TEF/BEF caused by esophageal diverticulum was rarely reported in the literatures. There were five case reports by Braghetto *et al.* (6), Tomiyama *et al.* (7), López *et al.* (3), Akashi *et al.* (8), and Stewart *et al.* (9) in available literatures researching (*Table 3*).

Based on medical history, clinical manifestations, radiographic examination, esophagoscopy and bronchoscopy, the diagnosis of TEF/BEFs or esophageal diverticula is not difficult (10). Esophagogram with oral taking contrast medium is helpful role in the exact localization of the fistula track. As the main methods to diagnose TEF/BEF, esophagoscopy and bronchoscopy are able to identify the fistulas in most cases. However, recognition of the tracheal or bronchial orifice in some smaller fistulas could be challenging (11). In our study, there were three cases with normal-appearing and one case with mucosa hypertrophy in bronchoscopy examination despite of obvious symptom with cough during swallowing. Orally administered methylene blue before bronchoscopy or observation of bubbles leaked from the airway may be helpful to identify small fistulas (11).

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Table 5 Keview of information on cases of esophagooroneman istura due to esophageat unverticulum in the interactives						
Author	Year	Cases	Location of fistula	Surgical approach	Interposed muscle	
Braghetto <i>et al.</i> (6)	2008	1	Middle of the esophagus-right main bronchus	EndoGIA and bronchial stapler was used to resect the diverticulum and the affected bronchus	Mediastinal pleural patch	
Tomiyama <i>et al.</i> (7)	2003	1	Middle of the esophagus- intermediate bronchus	Tract excision and both ends of fistula closed with ligation	N/A	
López <i>et al.</i> (3)	2003	1	Distal esophagus-branches of the lower left lobe	Diverticulum transection using mechanical sutures	Intercostal muscle	
Akashi <i>et al.</i> (8)	1997	1	Middle of the esophagus-right low lobe bronchus	Both diverticulum and fistulous closure to bronchus were divided with endo-stapler	Mediastinal pleural patch	
Stewart <i>et al.</i> (9)	1958	1	Middle of the esophagus- superior segment of right lower lobe	Segment resection and diverticulum excision	N/A	

Table 3 Review of information on cases of esophagobronchial fistula due to esophageal diverticulum in the literatures

The common symptoms of TEF/BEF include coughing while swallowing water (Ono's sign), fever, and/or weight loss. Continuous spillage of esophageal fluid into the tracheobronchial tree results in pneumonia, bronchial obstruction, atelectasis, and respiratory distress (12). Usually, repeated fever and poor food-in take lead to malnutrition of a patient. For patients with the Ono's sign associated pneumonia and infectious, the corrective procedure should be considered to close the fistulae with surgical management (13). However, for patients with small TEF/BEF that remain localized and symptomless, choosing active surgical intervention is still under debate. Devarbhavi *et al.* (14) documented that small TEF caused by esophageal tuberculosis can be treated safely with antituberculosis drugs alone.

Regarding surgical treatment for esophageal diverticula, the standard technique for TED is a long esophagomyotomy with diverticulectomy. For TED patients accompanied TEF/BEF, excision of the diverticulum and the fistulas with the adjacent inflammatory mass should be done. Employment of direct suture closure techniques of the tracheal/bronchus and esophageal defects with interposition of robust vascularized tissue is simple and sufficient (15). In our series, all patients fulfilled with limited diverticulectomy associated with tracheal/bronchus defects repair and the interposition of vascularized flaps, no recurrence fistula or symptomatic diverticula occurred during the follow-up period.

Isolated reports advocate initial management of nonmalignant TEF with endoscopic treatment, such as stent insertion or clip application. Jung *et al.* (16) described a successfully treatment of a quadriplegia patient with TEF through esophageal diverticulum by clipping under the esophagoduodenoscope on November 10, 2009. Schweigert et al. (17) also reported the recovery of three of four patients with benign gastro-tracheobronchial fistula treated by endoscopic stent insertion. Although endotherapy is a simple procedure for surgical application, controversy exists that endoscopic treatment could be definitively eliminated for acquired benign TEF. Buskens et al. (18) experienced failure in endoscopic treatment when attempting with a fibrin glue or hemostatic clips even in a patient with a long mediastinal fistula track. Muniappan et al. (2) advocated prompt surgical repair to control the fistula rather than depending on stents because stents may lead to significant granulation tissue and typically extend the airway injury. Bona et al. (19) recommended that endoscopic treatment should only be performed for patients with viable gastric tubes and no signs of mediastinitis or sepsis. To our best knowledge, we did not find similar literature on endoluminal stents in treating TEF through TED. The patients in our group were referred for surgical repair as a first choice after clear diagnosis. These nonsurgical invasive techniques may be of benefit for patients who decline or who are not candidates for surgery.

In summary, TEF/BEFs caused by traction diverticula is both rare and insidious. Early and definite surgical intervention is proposed once the diagnosis is confirmed. Single-stage limited diverticulectomy and direct suture repair of both the tracheal and esophageal defects with interposed muscle flaps can be performed successfully in TEF/BEFs patients caused by diverticulum.

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Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The study was approved by the Ethics Committee of Shanghai Chest Hospital [No. KS(Y) 1723] and written informed consent was obtained from all patients.

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