Name of Journal: World Journal of Gastrointestinal Surgery

Manuscript NO: 88922

Manuscript Type: CASE REPORT

Endoscopic treatment of extreme esophageal stenosis complicated with esophagotracheal fistula: A case report

Treatment of CES complicated with ETF

Jia-Heng Fang, Wei-Min Li, Cheng-Hai He, Jian-Liang Wu, Yun Guo, Zhi-Chao Lai, Guo-Dong Li

Abstract

BACKGROUND

Background: At present, there is no unified and effective treatment for extreme corrosive esophageal stenosis (CES) with esophagotracheal fistula (ETF). This case had extreme and severe esophageal stenosis (ES) and ETF after ingesting an enzyme-based chemical detergent, resulting in a serious pulmonary infection and severe malnutrition. Upper gastrointestinal imaging (UGI) showed that he had an ETF, and endoscopy showed he had extreme and severe esophageal stricture. This case was complex and was difficult to be treated. According to domestic and foreign literature, there is no universal treatment that is low-risk.

CASE SUMMARY

Recently, a patient came to our hospital with extreme ES, a ETF, and severe malnutrition complicated with pulmonary tuberculosis one month after the consumption of an enzyme-based detergent. The ES was serious, and the endoscope was unable to pass through the esophagus. We treated him with EIM, ESP and EBD by bronchoscope and gastroscope. This method not only closed the ETF, but also expanded...
the esophagus. **All treatment events are displayed on the timeline in Figure 1.** It solved the problem with minimal trauma and greatly reduced the pain of the patient. According to the literature and case reports at home and abroad, there are no similar cases.

**CONCLUSION**

We report, for the first time, a patient with extreme CES complicated with ETF, where the endoscope could not be passed through his esophagus but could be examined by bronchoscopy and treated by EIM, ESP and EBD.

**Key Words:** extreme CES; esophagotracheal fistula (ETF); endoscopic incision method (EIM); esophageal stent placement (ESP); endoscopic balloon dilation (EBD); Case report


**Core Tip:** Patients who have extreme CES with ETF often suffer a lot and have bad quality of their lives. We do not have clinical evidence-based guidelines, and there is great uncertainty regarding the best treatment approach. We report, for the first time, a patient with extreme CES complicated with ETF, where the endoscope could not be passed through his esophagus but could be examined by bronchoscopy and treated by EIM, ESP and EBD.

**INTRODUCTION**

Corrosive esophageal stenosis (CES) is rare but destructive; placing a significant burden on contemporary health systems around the world [1]. Patients who have extreme CES with esophagotracheal fistula (ETF) often suffer from typical chest pain,
severe pulmonary infection-related symptoms, malnutrition, etc., which seriously affect the quality of their lives [2]. Because of the rarity of this adverse event, clinicians usually have limited personal experience with it. We do not have clinical evidence-based guidelines, and there is great uncertainty regarding the best treatment approach.

For the treatment of extreme CES with ETF, there is no relevant guidelines or a relatively unified treatment plan in clinical practice. At present, the main clinical treatments for esophageal stenosis (ES) are the endoscopic incision method (EIM), endoscopic balloon dilation (EBD), gastroscope-assisted bougienage, esophageal stent placement (ESP) and local injection with glucocorticoids [3].

Recently, a patient came to our hospital with extreme ES, a ETF, and severe malnutrition complicated with pulmonary tuberculosis one month after the consumption of an enzyme-based detergent. The ES was serious, and the endoscope was unable to pass through the esophagus. We treated him with EIM, ESP and EBD by bronchoscope and gastroscope. This method not only closed the ETF, but also expanded the esophagus. All treatment events are displayed on the timeline in Figure 1. It solved the problem with minimal trauma and greatly reduced the pain of the patient. According to the literature and case reports at home and abroad, there are no similar cases.

**CASE PRESENTATION**

*Chief complaints*

The patient developed burning sensation and pain in the oral, throat, chest, behind the sternum and under the xiphoid process after taking enzyme detergent by mistake on May 5th, 2020.

*History of present illness*

None.

*History of past illness*
None.

**Personal and family history**
None.

**Physical examination**
Physical examination showed that the patient’s Watian drinking water experiment grade was a 5.

**Laboratory examinations**
The results are all shown in the manuscript by table.

**Imaging examinations**
The UGI, endoscopy results are all shown in the article by picture.

**MULTIDISCIPLINARY EXPERT CONSULTATION**
None.

**FINAL DIAGNOSIS**
extreme esophageal stenosis complicated with esophagotracheal fistula

**TREATMENT**
After we performed the endoscopic incision method (EIM) at the 19 cm narrow parts, we performed esophageal stent placement (ESP) at the upper esophagus and four times of endoscopic balloon dilations (EBDs) at the middle and lower esophagus. The stent was finally removed half and 4 mo after ESP.

**OUTCOME AND FOLLOW-UP**
After our careful treatment and a half reduction of fluid intake for one month, the patient's lung infection was significantly improved, and his weight increased by 10 kg. The symptoms of dysphagia were significantly improved, and the ETF was also sealed.

DISCUSSION

Since the physiological structure of the esophagus is thin and tubular, it is prone to stenosis when subjected to major injuries, such as corrosion and surgery. The incidence is approximately 1.1/100000 cases per year. Chronic ETF is extremely rare and occurs in approximately 3% of patients with chemically CES. The treatment mainly includes the repair of airway defects and esophageal reconstruction, which usually uses staging operation method. The first stage, as most clinicians think, is endoscopic esophageal dilatation (including EBD and bougienage). This case is very special for the following reasons: First, the patient is very young. If we choose surgical resection of the whole esophagus, the quality of life will be greatly reduced in the coming decades. Second, the patient’s endoscopy suggested that the esophagus was entirely narrowed with multiple fistulas. The thread guide was very likely to be displaced during the EBD. Even if the dilation were successful, the large balloon pressure could easily tear the fistula mucosa and cause further damage. Under our comprehensive consideration, we immediately performed EIM, which made it possible to expand the esophageal cavity and provide further ESP. As a new technology, EIM shows amazing feasibility and effectiveness.

There is no unified clinical treatment for complex CES. EIM is a new technology developed in recent years that was first used to treat recurrent Schatzki rings. Since 2012, it has gradually replaced EBD and is used to treat complex esophageal anastomotic stenosis. Wu et al. have shown that EIM for complex ES is safe and can significantly alleviate the clinical symptoms of dysphagia in a short time, but its long-term effect is still uncertain. Li, Hordijk et al. believe that EIM benefits patients who have severe and complex benign ES more and have fewer side effects. In this case, the total esophageal tube was narrow, there were multiple sinuses in the esophagus, and it was difficult to reach the gastric cavity with the guidewire. We performed EIM...
for upper ES, which expanded the esophageal cavity and created the possibility of follow-up treatment.

As one of the treatments for ES, ESP has many adverse events, such as chest pain, stent displacement or shedding, and tissue embedding stents. Fuccio et al.\(^{[12]}\) performed a meta-analysis that indicated that approximately 28.6% of patients had esophageal stent displacement and 20.6% had adverse events. Therefore, ESP is not recommended as the best treatment for benign ES\(^{[3]}\). For cases of malignant ES complicated with ETF, ESP is the recommended scheme\(^{[3]}\). For the treatment of benign ES combined with ETF, a unified recommended scheme is void. The patient faced the following two problems on admission: 1. The patient's esophagus was completely narrow, and the endoscope was unable to pass through; 2. The patient had stenosis complicated with an ETF. Therefore, we believe that the most appropriate method is EIM and ESP. On the one hand, the covered stent can block the ETF and promote self-healing; on the other hand, the covered stent can dilate the narrow esophagus and alleviate the symptoms of dysphagia. There were no adverse event, such as stent displacement or stent insertion. The esophagus was unobstructed after placement, and the fistula was closed after stent removal. It was proven to be a reasonable treatment for benign ES complicated with ETF.

Patients with extreme CES often have to accept EBD dozens of times. They go through a great deal, and the esophagus is easy to tear after operation. The medical cost is also high. The expansion success rate of corrosive stenosis is approximately 50%, which is significantly lower than that of other benign stenoses (75-80%)\(^{[7]}\). For the stenosis part located below the esophageal stent in this case, considering the tear caused by ordinary balloon dilatation, we used the duodenal papilla columnar expansion balloon to expand the esophagus by dividing segments. We performed this three additional times at 7, 21 and 35 days after the initial EBD visit. The inner diameter of the balloon increased in turn during each expansion. The patient maintained a liquid and semiliquid diet during this period, without obvious tearing, perforation, bleeding or other adverse events. For the treatment of lower ES, there are two choices; ESP or ESP
after EBD. More clinical studies are needed to prove which one has better safety, efficacy and economic benefits.

For the treatment of benign ES, Xie\textsuperscript{[13]} et al. believe that oral administration of glucocorticoids in patients with diabetes, hypertension, infection, tuberculosis and other patients will lead to aggravation, so it is not recommended. The effect of glucocorticoid injection is ideal, but no drug injection has been proven to be effective for the treatment of chemically CES, and corticosteroids also have no benefits\textsuperscript{[14]}. Enzyme-based chemical detergent can etch patient's esophagus, long-standing tuberculosis infection of more than 2 months also could lead to an acquired ETF. We believe that ATT treatment is also crucial for the healing of ETF and subsequent recovery, so we used ATT immediately as soon as tuberculosis was confirmed. Khan et.al\textsuperscript{[15]} cured a patient who had tuberculous fistulas successfully with ATT, supports our view.

Chemical corrosive esophageal injury should be examined by gastroscopy combined with UGI, nasal endoscopy and even bronchoscopy when necessary. For treatment, we should personally select EIM, EBD, and ESP according to every patient's specific situation. Due to entire CES combined with tracheoesophageal fistula having a wide range of stenosis, the treatment is extremely difficult. Clinical treatment should be taken under individualized assessment, and combined treatment should be given when necessary. In this case, we used a bronchoscope with a thinner diameter to evaluate and treat ES repeatedly instead of a gastroscope, which is rare in the clinic and has a perfect effect. Whether the ETF and ES will recur in this patient remains to be observed. In the future, we need more treatment research data to guide treatment.

\textbf{CONCLUSION}
We report, for the first time, a patient with extreme CES complicated with ETF, where the endoscope could not be passed through his esophagus but could be examined by bronchoscopy and treated by EIM, ESP and EBD.
Peiyao Wu, Fei Wang, Xiaochao Wu, Junjie Nie, Xianxiu Ge, Quanpeng Li, Jie Lin, Lin Miao. "Comparison of esophageal stent placement versus endoscopic incision method for treatment of refractory esophageal anastomotic stricture", Annals of Palliative Medicine, 2019