

Choledochoscope-assisted antegrade/retrograde endoscopic dilation of complete esophageal stenosis in a patient with dystrophic epidermolysis bullosa

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Abstract

Dystrophic epidermolysis bullosa is a mucocutaneous disorder, characterized by recurrent formation of blisters and scarring. The gastrointestinal tract is commonly affected by the disease and the proximal esophagus is the most common area of involvement of the gastrointestinal tract. The esophageal strictures are common in patients with dystrophic epidermolysis bullosa that can lead to complete esophageal stenosis in some cases. The antegrade/retrograde endoscopic dilation is a commonly used method in these patients. Different kinds of endoscopes may be used for the retrograde procedure, such as conventional upper gastrointestinal (UGI) endoscopes, slim-paediatric UGI endoscopes and ultrathin UGI nasal endoscopes. Herein, we reported the first antegrade/retrograde esophageal dilation case performed under choledochoscopic guidance. (Acta gastroenterol. belg., 2020, 83, 491-492).

Keywords : dystrophic epidermolysis bullosa, esophageal stenosis, esophageal dilation, choledochoscope.

Introduction

Dystrophic epidermolysis bullosa (DEB) is a rare inherited disorder, characterized by recurrent formation of blisters and scarring induced by minor mechanical trauma. Mutations of the type 7 collagen gene is the underlying cause of disease and the areas where type 7 collagen is expressed such as skin, oral mucosa, gastrointestinal tract and genitourinary tract are affected by the disease (1). The proximal esophagus is the most common area of involvement of the gastrointestinal tract and the minor traumas caused by swallowing solid foods leads to blister formation and scars that result in esophageal strictures. The esophageal strictures are common in patients with DEB and have been reported in 70%-87% of cases. This can lead to complete esophageal stenosis in some cases (1,2). The antegrade/retrograde endoscopic dilation of the esophagus is a commonly used method in these patients (1-3). In this manuscript, we presented the first antegrade/retrograde esophageal dilation case performed under choledochoscopic guidance.

Case Report

A 27-year-old male with dystrophic epidermolysis bullosa was referred to our clinic for worsening dysphagia. The patient had a history of antegrade esophageal dilation many times for epidermolysis bullosa-related esophageal

stenosis and enteral feeding had been supported with an existing percutaneous endoscopic gastrostomy (PEG) tube for 12 years. Endoscopic examination revealed complete stenosis in the post-cricoid area of the esophagus and further attempts for antegrade dilation failed due to non-passage of the guidewire through the occlusion. A contrast esophagram was not performed on the patient due to the aspiration risk. On the following day, a choledochoscope-assisted antegrade/retrograde esophageal dilation procedure was performed on the patient under general anesthesia. The procedure steps were as follows; A conventional fibercholedochoscope (FCP-9P Choledochofiberscope, Pentax Medical) was passed through the existing PEG tube (Corflo-cuBBY, 20FR, 2cm, Halyard Health) into the stomach. The choledochoscope was then directed towards the gastroesophageal junction and passed into the proximal esophagus with direct visualization by the endoscopist (Figure-1A/1B). A guidewire (Jagwire, 0.035 in, 450 cm, Boston Scientific) was pushed retrograde towards the upper esophagus via the working channel of the choledochoscope and was passed through the stenosis area of the esophagus under fluoroscopic guidance (Figure-1C). A flexible endoscope was advanced antegradely and the guidewire, which was identified in the hypopharynx, was grasped and pulled out of the mouth using endoscopic forceps (Figure-1D). The bougie dilators were passed over the guidewire, through the peroral route and esophageal dilatation was performed sequentially with 15-30 FR bougies. The oral intake of the patient improved after the dilatation procedure and he was discharged from the hospital with no complications.

Discussion

The antegrade/retrograde endoscopic dilation of the esophagus is a commonly used method in patients with complete esophageal stenosis that may occur in

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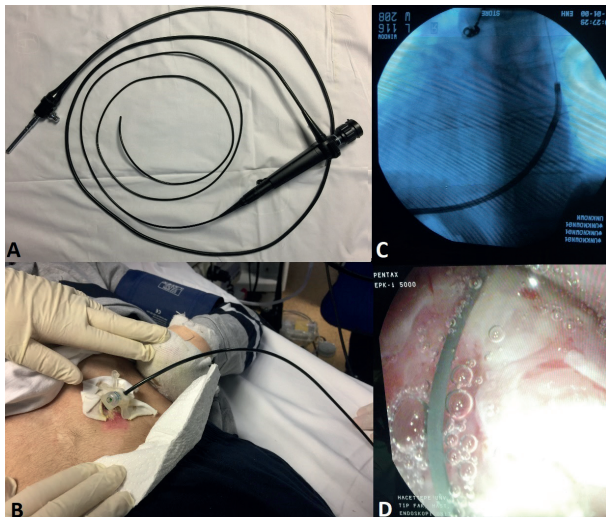


Figure 1. — A : The choledoscope image used in the procedure. B-D : The steps of the procedure; A conventional fibercholedoscope was passed through the PEG tube into the stomach and was passed into the proximal esophagus with direct visualization by the endoscopist (B). A guidewire was passed through the stenosis area of the esophagus under fluoroscopic guidance (C). A flexible endoscope was advanced antegradely and the guidewire, which was identified in the hypopharynx, was grasped and pulled out of the mouth using endoscopic forceps (D). The bougie dilators were passed over the guidewire, through the peroral route and esophageal dilatation was performed.

the course of various diseases including DEB (3-7). In this method, one scopic device is pushed forward through the oral route antegradely, and the second one is pushed forward through the gastrostomy (GT) site retrogradely, towards the esophageal stenosis area (3-6). Different kinds of endoscopes may be used for the retrograde procedure, such as conventional upper gastrointestinal (UGI) endoscopes, slim-paediatric UGI endoscopes and ultrathin UGI nasal endoscopes. Depending on the diameter of the scopic device used for the dilation procedure, removal of the PEG tube, dilation of the gastrostomy site and insertion of a new PEG tube after the dilation may be required. There are some complications of dilation of the gastrostomy site, such as separation of the stomach from the abdominal wall and pneumoperitoneum, GT site leak, GT site infection, and loss of GT track. Furthermore, the removal of the PEG tube before the procedure and insertion of a new one after

the procedure has an additional cost (3-6). In the current case, a fibercholedoscope was used for the retrograde route. To the best of our knowledge, our experience is the first case of antegrade/retrograde esophageal dilation performed under choledoscopic guidance. The noteworthy advantage of using a choledoscope for this procedure is primarily associated with the ultra-thin diameter of the device (insertion tube outer diameter 3.1 mm) that allows it to pass through PEG tubes. With this method, removal of the PEG tube before the procedure and dilation of the gastrostomy site is not necessary. Therefore, dilation-related complications do not occur and insertion of a new PEG tube is not needed in these patients.

Consequently, antegrade/retrograde esophageal dilation can be performed under choledoscopic guidance in patients with complete esophageal stenosis. Using a choledoscope for dilation procedure has noteworthy advantages compared to using conventional UGI endoscopes about the procedure-related complications and costs.

Conflict of interests

All authors declare that they have no conflict of interest.

References

1. SPILIOPOULOS S., SABHARWAL T., KROKIDIS M., GKOUTZIOS P., MELLERIO J., DOURADO R. *et al.* Fluoroscopically guided dilation of esophageal strictures in patients with dystrophic epidermolysis bullosa : long-term results. *AJR Am J Roentgenol.*, 2012, **199** : 208-12.
2. CASTILLO R., DAVIESYK., LINYC., GARCIA M., YOUNG H. Management of esophageal strictures in children with recessive dystrophic epidermolysis bullosa. *J. Pediatr. Gastroenterol. Nutr.*, 2002, **34** : 535-41.
3. ANDERSON BT., FEINSTEIN JA., KRAMER RE., NARKEWICZ MR., BRUCKNER AL., BRUMBAUGH DE. The approach and safety of esophageal dilation for treatment of strictures in children with epidermolysis bullosa. *J. Pediatr. Gastroenterol. Nutr.*, 2018, **67** : 701-705.
4. GOGUEN LA, NORRIS CM., JAKLITSCH MT., SULLIVAN CA., POSNER MR., HADDAD RI. *et al.* Combined antegrade and retrograde esophageal dilation for head and neck cancer-related complete esophageal stenosis. *Laryngoscope*, 2010, **120** : 261-6.
5. GAVRIEL H., DUONG C., SPILLANE J., SIZELAND A. Bidirectional esophageal dilatation in pharyngo-esophageal stenosis postradiotherapy. *Head Neck*, 2013, **35** : 733-7.
6. BERTOLINI R., MEYENBERGER C., PUTORA PM., ALBRECHT F., BROGLIE MA., STOECKLI SJ. Endoscopic dilation of complete oesophageal obstructions with a combined antegrade-retrograde rendezvous technique. *World J. Gastroenterol.*, 2016, **22** : 2366-72.
7. ILKIN NAHARCI M., TUZUN A., ERDIL A., ATEŞ Y., BAGCI S., YAMANEL L. *et al.* Effectiveness of bougie dilation for the management of corrosive esophageal strictures. *Acta Gastroenterol. Belg.*, 2006, **69** : 372-6.