International Journal of Pediatric Otorhinolaryngology 98 (2017) 158–161

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: http://www.ijporlonline.com/

Review Article

Endoscopic esophageal and tracheal cauterization for closure of recurrent tracheoesophageal fistula: A case report and review of the literature

Stacie Gregory, MD [a, *], Robert H. Chun, MD [a], Daiva Parakininkas, MD [c], Louella Amos, MD [c], Roger Fons, MD [d], Diana G. Lerner, MD [b], Dave R. Lal, MD MPH [e], Cecille Sulman, MD [a]

[a] Department of Otolaryngology, Medical College of Wisconsin (MCW), Milwaukee, WI, United States
[b] Department of Pediatrics, Division of Gastroenterology, Hepatology and Nutrition, MCW, Milwaukee, WI, United States
[c] Department of Pediatrics, Division of Pulmonary Medicine, MCW, Milwaukee, WI, United States
[d] Department of Anesthesiology, MCW, Milwaukee, WI, United States
[e] Department of Surgery, Division of Pediatric Surgery, MCW, Milwaukee, WI, United States

ARTICLE INFO

Article history:
Received 14 March 2017
Received in revised form
27 April 2017
Accepted 30 April 2017
Available online 2 May 2017

Keywords:
Endoscopic
Recurrent
Tracheoesophageal fistula
TEF
Electrocautery
Treatment

ABSTRACT

Objective: Recurrent tracheoesophageal fistula (TEF) can be a diagnostic and therapeutic challenge. Traditional treatment is thoracotomy, which carries significant morbidity and technical difficulty especially in a previously operated field. Recently, endoscopic techniques have been advocated as a primary approach for treatment of recurrent TEF prior to open repair. This case report describes the endoscopic technique used to address a recurrent TEF. The existing literature of all reported endoscopic cauterization methods is reviewed.

Methods: An 8 month old with proximal esophageal atresia and distal TEF underwent endoscopic closure of a recurrent TEF. The fistula was approached endotracheally utilizing Bugbee electrocautery (EC) and endoluminally through the esophagus using argon plasma coagulator and placement of porcine submucosa graft into the tract. Current literature review is presented with a synthesis of data on cases utilizing endoscopically applied EC and the combined results of this closure technique.

Results: Our patient has maintained successful closure after a single treatment confirmed on follow up endoscopy 6 months post repair. Including this patient, there have been 30 patients with recurrent TEF treated utilizing endoscopic EC reported in the literature. **The overall success rate is 78.8% with a mean of 1.88 procedures per successful closure.** Comparing EC alone to EC combined with tissue glues or laser, success rates are **67% and 86%** respectively.

Conclusion: Endoscopic repair of recurrent TEF has proven to be safe and effective in the literature as an alternative to a second thoracotomy/open surgical repair. EC combined with tissue glues or laser is more effective than EC alone based on available data.

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* Corresponding author. N91W16015 Junction Way Apt 106, Menomonee Falls, WI 53051, United States.
E-mail addresses: srgregory@siumed.edu, srgregory@mcw.edu (S. Gregory), rchun@mcw.edu (R.H. Chun), dparakin@mcw.edu (D. Parakininkas), lamos@mcw.edu (L. Amos), rfons@mcw.edu (R. Fons), dglerner@mcw.edu (D.G. Lerner), dlal@mcw.edu (D.R. Lal), csulman@mcw.edu (C. Sulman).

http://dx.doi.org/10.1016/j.ijporl.2017.04.051
0165-5876/

1. Introduction

After surgical repair of esophageal atresia (EA) with tracheoesophageal fistula (TEF), recurrence of the TEF occurs at a rate of 5–10% [1–3]. Treatment of recurrent TEF can be quite difficult and traditionally was approached by thoracotomy. Given the significant potential morbidity as well as technical difficulty encountered in a previously operated field, an interest in less invasive treatment options has arisen. Endoscopic techniques are now being advocated as an alternative to open repair. Endoscopic approaches are taken from the tracheal and or esophageal side and range from the use of electrocautery (EC), laser, or wire brushing to denude the epithelial tract and promote scarring, to applications of tissue glues or vascularized pieces of tissue to occlude the tract. Often times, a combination of techniques is utilized.

We encountered an 8 month old with recurrent TEF who we elected to repair endoscopically, combining treatment modalities of argon plasma coagulator and EC, addressing both the esophageal and tracheal side of the fistula respectively. Due to the immediate and lasting success of this procedure, we reviewed the available literature to determine the overall success rates of endoscopic closure of recurrent TEF when using EC techniques.

2. Materials and methods

We will describe a case of recurrent congenital TEF treated at Children's Hospital of Wisconsin with combined endoscopic approaches from both the tracheal and esophageal side utilizing EC as well as argon plasma coagulator and Surgisis (Cook Medical, Bloomington, IN).

A literature review of the PubMed Database was performed to identify all available English language literature pertaining to the endoscopic repair of recurrent TEF using EC methods.

2.1. Case report

The patient had a history of premature birth (34 weeks gestation) with persistent desaturations following delivery. Workup revealed proximal esophageal atresia with a distal TEF. The patient was transferred to the Children's Hospital of Wisconsin for further management. Further assessment revealed vascular compression of esophagus by an aberrant right subclavian artery arising from the descending aorta as well as coarctation of the aorta.

The patient underwent primary repair of the EA/TEF at 3 weeks of age. The post operative course was complicated by a contained leak at the anastomosis that resolved with conservative management by 5 weeks of age. The patient was discharged to home at 2 months on an oral diet.

At 3 months of age, the patient underwent an elective anastomotic balloon dilation by Interventional Radiology. After the procedure, he had significant respiratory distress and recurrent apparent life threatening events (ALTE's), especially prominent during agitation and crying. Record review of the month prior to this did not reveal any documented complaints of respiratory distress, coughing with feeds or dysphagia. **Bronchoscopy found 80% narrowing of the distal trachea secondary to extrinsic compression**. CT with angiogram confirmed these findings. At 3.5 months of age, the patient underwent **aortopexy with innominate artery suspension**. He was discharged home on POD 11 with marked improvement and no further cyanotic episodes or respiratory distress.

At multidisciplinary Aerodigestive and Foregut Clinic (ADFC) follow up appointment at 4 months of age, the mother felt the patient was doing well and having no symptoms of cough or respiratory distress. However, he returned to ADFC at 8 months of age with complaints of recurrent cough and frequent respiratory illnesses for the past 2 months. Coughing was noted with feeds but no nocturnal cough or coughing with activity. Triple endoscopy (direct laryngoscopy, bronchoscopy and esophagoscopy) was performed the following week. The flexible bronchoscopy revealed recurrence of the TEF, demonstrated by using 1 cc of diluted methylene blue (0.1 cc in 10 cc of saline) injected into the residual tracheoesophageal pouch using a #5 flexible suction catheter. Flexible esophagoscopy was performed immediately following. Blue dye was seen at the site of the anastomosis, confirming recurrent TEF (Fig. 1).

Closure of the recurrent TEF was carried out first on the esophageal side, using **Argon plasma coagulator** to burn the small defect. Next, a 3 mm **Bugbee electrocautery device** inserted through the side port of a 3.5 rigid bronchoscope, was used to cauterize (coagulation setting of 10) the residual tract on the tracheal side (Fig. 2). A dry 8 ply **Surgisis (0.5 cm) was placed with a telescopic forceps in a parachute fashion** and applied on the tracheal side of the fistula. No tissue glue was placed. There were no complications following the procedure.

Return to the OR at 3 and 6 months post repair to reassess closure of the TEF with methylene blue test confirmed successful repair; follow up appointment at 13 months of age revealed no complaints of cough or aspiration and no respiratory illnesses.

3. Results

We identified 10 studies describing the use of endoscopically applied EC in treatment of recurrent TEF (Table 1). Studies comprised 31 patients (32 fistulas) in all. With the addition of our single case, data analysis was carried out on 32 patients (33 fistulas). Successful closure was seen in 26 of 33 fistulas (78.8%). Of these, only 12 were treated with EC alone. The remaining 20 were

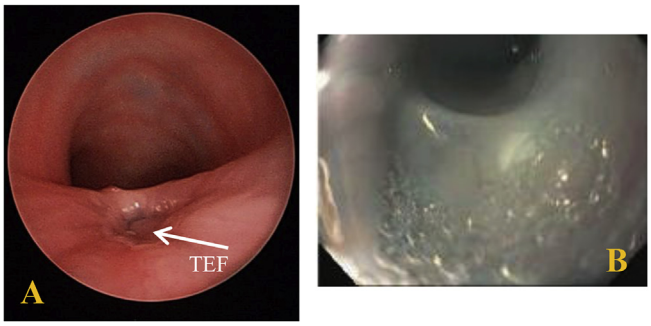

Fig. 1. A: Bronchoscopic image of the recurrent TEF. B: Methylene blue dye in the esophagus after it was instilled from the trachea into the fistula tract.