Accepted manuscripts are the articles in press that have been peer reviewed and accepted for publication by the Editorial Board of the *Vojnosanitetski Pregled*. They have not yet been copy edited and/or formatted in the publication house style, and the text could still be changed before final publication.

Although accepted manuscripts do not yet have all bibliographic details available, they can already be cited using the year of online publication and the DOI, as follows: article title, the author(s), publication (year), the DOI.

Please cite this article **SURGICAL TREATMENT OF ACQUIRED TRACHEOESOPHAGEAL FISTULA CAUSED BY BALLOON DILATATION OF CORROSIVE ESOPHAGEAL STRicture IN A CHILD**

**HIRURŠKO LEČENJE STEČENE TRAHEOEZOFAGUSNE FISTULE PROUZROKOVANE BALON DILATACIJOM KOROZIVNE STENOZE JEDNJAVA KOD DETETA**

Authors Dejan Stojakov*§, Maja Miličković†§, Predrag Minić ‡§, Miroslav Vukadin †, Nikola Stanković ||§, Đorđe Savić †§, Vojnosanitetski pregled (2018); Online First October, 2018.

UDC:

DOI: [https://doi.org/10.2298/VSP180615169S](https://doi.org/10.2298/VSP180615169S)

When the final article is assigned to volumes/issues of the Journal, the Article in Press version will be removed and the final version appear in the associated published volumes/issues of the Journal. The date the article was made available online first will be carried over.
SURGICAL TREATMENT OF ACQUIRED TRACHEOESOPHAGEAL FISTULA CAUSED BY BALLOON DILATATION OF CORROSIVE ESOPHAGEAL STRicture IN A CHILD

HIRURŠKO LEČENJE STEČENE TRAHEOEZOFAGUSNE FISTULE PROUZROKOVANE BALON DILATACIJOM KOROZIVNE STENOZE JEDNJAKA KOD DETETA

Dejan Stojakov*§, Maja Miličković†‡§, Predrag Minić‡§, Miroslav Vukadin †, Nikola Stanković||§, Đorđe Savić †§

*Clinical Center of Serbia, Clinic for Digestive Surgery- First Surgical University Hospital, Department of Esophagogastric Surgery, Belgrade, Serbia; Mother And Child Health Care Institute of Serbia “Dr. VukanČupić”, † Department of Abdominal Surgery, ‡ Department of Pulmonology, || Department of Anesthesiology and Intensive Therapy, Belgrade, Serbia; § School of Medicine, University of Belgrade, Belgrade, Serbia

Corresponding author and person to whom requests for separates can be sent:
Maja Milickovic
Department of Abdominal Surgery, Mother and Child Health Care Institute of Serbia, 6-8 Radoja Dakica St., 11070 Belgrade, Serbia. Tel.: +381113108183, +38163335290
e-mail: majacvetkovicmilickovic@gmail.com

Running title: Acquired tracheoesophageal fistula

Criteria for inclusion in the authors'/ contributors’ list:


Statement that the manuscript has been read and approved by all the authors/contributors, that the requirements for authorship have been met, and that each author believes that the manuscript represents honest work.

1. Dejan Stojakov
2. Maja Miličković
3. Predrag Minić
4. Miroslav Vukadin
5. Nikola Stanković
6. Đorđe Savić
Abstract

**Introduction.** Tracheoesophageal fistula (TEF) as a complication of balloon dilatation (BD) of corrosive esophageal stricture is a very rare and serious condition. Life threatening aspiration pneumonia requests urgent lungs' protection, but overall treatment strategy is not clearly defined. **Case report.** Twenty-month-old female child accidentally ingested a household bleach. Caustic injury of esophagus was healing with development of strictures of cervical and proximal thoracic esophagus. TEF was developed during the third BD. Healing of TEF and pulmonary infection was achieved by exclusion of the esophagus (pharyngostomy and feeding gastrostomy) together with prolonged tracheobronchial intubation and toilette. Retrosternal colon interposition was performed a year later, with excellent functional results over four-year follow-up. **Conclusion.** Esophageal exclusion in the first stage, and pharyngoesophageal reconstruction in the second stage, is a useful therapeutic option in the treatment of TEF caused by balloon dilatation of corrosive esophageal stricture in children.

Key words: corrosive esophageal stricture, balloon dilatation, acquired tracheoesophageal fistula, reconstruction of the esophagus.

Introduction

Corrosive agents ingestion in children is accidental and usually caused by alkalies. Deep tissue involvement with inflammatory process often causes esophageal stricture which requires dilatation or even esophageal replacement. Esophageal perforation with formation of tracheoesophageal fistula (TEF) during dilatation of corrosive esophageal stricture is a very rare complication, and may lead to fatal aspiration pneumonia.[1,2,4]
There is no consensus about the treatment, and we believe that each case report can help in outlining the optimal strategy for distinct clinical presentations.

Case report

Twenty-month-old female child accidentally ingested a household bleach containing sodium hydroxide. Caustic injury of esophagus was healing with development of strictures of cervical and proximal thoracic esophagus. Barium swallow showed stenosis of the cervical and proximal thoracic part of the esophagus without stenosis of infracarinal part of esophagus and stomach, and without gastroesophageal reflux. There were no clinical signs of inhalation injury. Balloon dilatations started 15 days after the injury, in ten-day intervals. First two dilatations were performed without complications and sufficient peroral feeding was possible between them. Hypersalivation and coughing appeared immediately after the third dilatation. The next day there was a clinical deterioration manifested by tachypnea, tachycardia and high body temperature, accompanied by leukocytosis. A chest X-ray showed bilateral infiltrates in basal lung zones without mediastinal widening. Fiberoptic tracheobronchoscopy showed a 15 mm long laceration of the membranous tracheal wall, located about 1 cm above the carina (Figure 1a). Cervicothoracic CT scan, after pulling upward sof endotracheal tube, confirmed the 15 mm long fistula between the strictured esophagus and the membranous tracheal wall (Figure 2). There were bilateral lung infiltrates, without gas and liquid collection in the mediastinum.

We opted for two-stage surgery. First stage consisted of esophageal exclusion with forming of salivary fistula and gastrostomy (Figure 3). Prolonged tracheal intubation with tracheobronchial toiletwas planned in the postoperative period, expecting spontaneous healing of the TEF (Figure 4). During the surgery through the left cervicotomy, the cervical esophagus firmly attached to the trachea was found, without possibility of encircling it. Lateral pharyngotomy was performed, stenotic cervical esophagus was completely obliterated by several individual stitches, and pharyngostomy was created (Figure 5). The pulmonary infection was gradually cured postoperatively. Tracheobronchial fiberoptic endoscopy was used to treat atelectasis of the left lung. On the 10th postoperative day, since the bronchoscopy showed healed laceration on membranous tracheal wall, the child was extubated (Figure 1b). The child was fed through gastrostomy, without signs of gastroesophageal reflux.

The second stage surgery, bypass retrosternal colon interposition with pharyngocolic anastomosis, was performed a year later. Before reconstruction, we confirmed normal vocal cord function, good pulmonary function, and normal nutritive status. We used long isoperistaltic colonic segment, vascularized by left colic vessels (Figure 6).

Over four-year follow-up, the girl was normally fed perorally, with normal development and without occurrence of pulmonary infections (Figure 7).

Discussion

Tracheoesophageal fistula occurring during pneumatic dilatation of esophageal corrosive stricture is a very rare complication and a great challenge for clinicians. According to our best knowledge, only 10 similar cases were reported until now. With no clearly defined treatment strategy, the approach should be individually tailored, taking into account all relevant clinical features. Anatomy of esophageal stenosis, localization and appearance
of the TEF, the presence of pulmonary, mediastinal, and pleural infection and signs of sepsis, age, nutritional status and general condition of the patient, together with operative findings are essential for treatment strategy.\(^{[3,4]}\)

Symptoms of the TEF may be different. The slightest suspicion of the TEF requires timely and adequate diagnostic procedure. Esophagography and esophagoscopy can show anatomy of the TEF.\(^{[5]}\) Bronchoscopy allows more accurate localization and assessment of the TEF morphology.\(^{[5]}\) CT scan gives information about inflammatory changes in the mediastinum, pleural cavity and the lungs, but details of TEF anatomy can remain undetected in intubated patients.\(^{[5,8]}\) According to our experience in this case, we believe that CT examination may be more useful when endotracheal tube is slightly pulled upward to the larynx to express TEF (Figure 2). CT may show localization and size of TEF, and allow precise surgical planning, too.

The violation of the membranous wall of the trachea during BD of the corrosive esophageal is the result of transmural inflammation. Adhesions between the wall of the esophagus and the membranous wall of the trachea essentially make the walls of these organs behave as a single rigid structure. Fibrosis involving both esophageal and membranous tracheal wall complicates the primary surgical reparation of the TEF, but may help in spontaneous healing of the TEF (Figure 4). Lungs protection from aspirated saliva and refluxed gastric content contamination is crucial for the treatment of TEF. Treatment with esophageal stenting as bridging procedure before definitive surgical treatment may be useful, but can be controversial since esophageal stent covers the TEF, but separates the edges of the TEF and does not support the healing.\(^{[5]}\) Surgical exclusion of the esophagus is another way to protect lungs. In both cases, delayed reconstruction of the esophagus is the second stage of the treatment.\(^{[5]}\) Various endoscopic options such as glue, laser, and cauterization are in use for smaller TEF of some other etiology, while primary surgical TEF repair is adequate only when it is not too risky for airway safety.\(^{[3,6]}\)

The staged surgical treatment is an optimal solution for the TEF caused by the dilatation of a corrosive esophageal stenosis. Our patient had a similar treatment as a young adult with caustic ingestion reported in the literature.\(^{[9]}\) In our case, the primary repair of the TEF was risky. Effective protection of the lungs and the spontaneous healing of the TEF was achieved by exclusion of the esophagus, together with repeated tracheobronchial toiletté through the endotracheal tube. Delayed retrosternal colonic interposition with pharyngocolic anastomosis was performed according to our previous experience.\(^{[10]}\) Over four-year follow-up excellent functional results were achieved.

**Conclusion**

Two-stage surgical treatment consisting of esophageal exclusion esophagostomy or pharyngostomy and gastrostomy in the first stage, and pharyngoesophageal reconstruction with retrosternal colonic interposition in the second stage, is a useful therapeutic option in the treatment of tracheoesophageal fistula caused by pneumatic dilatation of corrosive esophageal stricture in children.

**Acknowledgement**

Acknowledgement to Professor Radoslav Jakovic, thoracic surgeon (departed), School of Medicine, University of Belgrade; Institute for Lung Diseases, Thoracic Surgery and
Tuberculosis, Clinical Center of Serbia, for his great support in determining therapeutic strategy for our patient.

References


Figure 1.
Figure 2.

Figure 3.

Figure 4.
Legends for illustrations

Figure 1: Bronchoscopic appearance of TEF initially (a) and ten days after surgery (b)

Figure 2: CT findings after pulling upwards endotracheal tube, sagittal (a) and axial plane (b)(TEF - black arrow)

Figure 3: (a) TEF and (b) surgical procedure (schema)
Figure 4: Mechanism of TEF development caused by pneumatic dilatation (a, b, c) and spontaneous healing of TEF(d) (schema)

Figure 5: Exclusion of the stenotic cervical esophagus (a) and lateral pharyngostomy (b)

Figure 6: Temporary clamping of middle colic vessels and marginal vascular arcade (a) and isoperistaltic colonic conduit vascularized by left colic artery (b)

Figure 7: Plain (a) and lateral (b) contrast radiographies two weeks after pharyngoesophageal reconstruction with retrosternal colon interposition

Received on Juny 15, 2018.
Revised on October 8, 2018.
Accepted on October 11, 2018.
Online First October, 2018.