CASE REPORT

UDC: 616-053.2:616.329-001.37-089
https://doi.org/10.2298/VSP180615169S

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[†‖], Djordje Savić[†‡]

Clinical Center of Serbia, First Surgical University Hospital, Clinic for Digestive Surgery *Department of Esophagogastric Surgery, Belgrade, Serbia; University of Belgrade, [†]Faculty of Medicine, Belgrade, Serbia; Mother and Child Health Care Institute of Serbia "Dr. Vukan Čupić", [‡]Department of Abdominal Surgery, [§]Department of Pulmonology, [‖]Department of Anesthesiology and Intensive Therapy, Belgrade, Serbia

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 (pharyngostoma 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:
esophageal stenosis; burns, chemicals; dilatation; tracheoesophageal fistula; digestive system surgical procedures; child preschool; treatment outcome.

Apstrakt

Uvod. Traheoezofagusna fistula (TEF), kao komplikacija balon dilatacije (BD) korozivne stenoze jednjaka, je vrlo retko i teško stanje. Po život opasna aspiraciona pneumonija zahteva urgentnu zaštitu pluća, ali opšta strategija lečenja nije jasno definisana. **Prikaz bolesnika.** Dvadesetomesečna devojčica je zadesno progutala kućni izbeljivač. Stenoza vratnog i proksimalnog grudnog jednjaka razvila se kao posledica kaustične povrede jednjaka. Do razvoja TEF došlo je tokom treće procedure BD. Sanacija TEF i plućne infekcije postignuta je ekskluzijom jednjaka (faringostoma i nutrutivna gastrostoma) uz prolongiranu traheobronhijalnu intubaciju i toaletu. Retrosternalna interpozicija dugog segmenta kolona je urađena nakon godinu dana, sa odličnim funkcionalnim rezultatom tokom četvorogodišnjeg praćenja. **Zaključak.** Ekskluzija jednjaka u prvom, i faringoezofagealna rekonstrukcija u drugom stadijumu, predstavlja efikasan način lečenjaTEF nastale prilikom BD korozivne stenoze jednjaka.

Ključne reči:
jednjak, stenoza; opekotine hemijskim sredstvima; dilatacija; fistula, traheoezofagusna; hirurgija, digestivnog sistema, procedure; deca, predškolska, lečenje, ishod.

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 [1–4].

Correspondence to: Maja Miličković, Department of Abdominal Surgery, Mother and Child Health Care Institute of Serbia "Dr. Vukan Čupić", 6-8 Radoja Dakića St., 11070 Belgrade, Serbia. E-mail: majacvetkovicmilickovic@gmail.com

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. Ballon 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 computed tomography (CT) scan, after pulling upwards of 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.

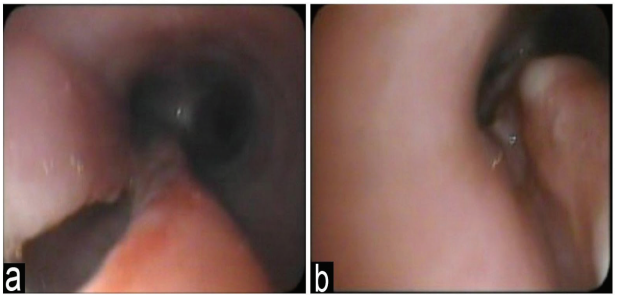

Fig. 1 – Bronchoscopic appearance of tracheoesophageal fistula (TEF) initially (a) and ten days after surgery (b).

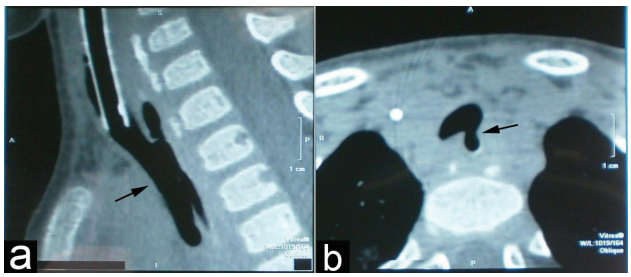

Fig. 2 – Computed tomography (CT) findings after pulling upwards endotracheal tube, sagittal (a) and axial plane (b) (TEF – black arrow).

We opted for two-stage surgery. First stage consisted of esophageal exclusion with forming of salivary fistula and gastrostomy (Figure 3).

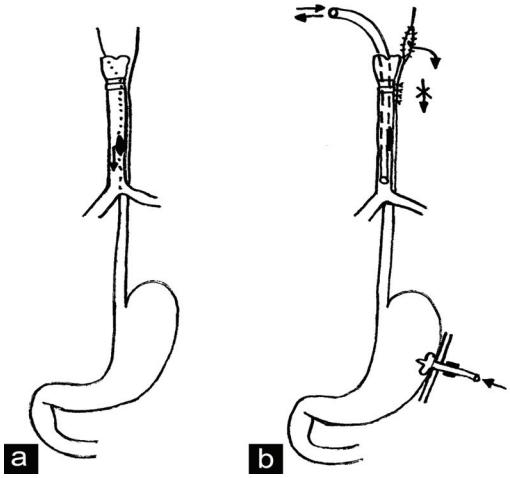

Fig. 3 – Schematic presentation of tracheoesophageal fistula (TEF) (a) and surgical procedure (b).

Prolonged tracheal intubation with tracheobronchial toilet was 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 open circling it.

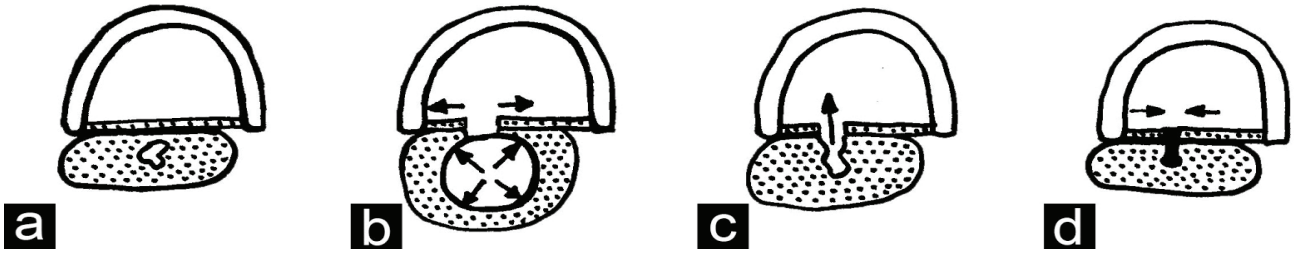

Fig. 4 – Schematic presentation of the mechanism of tracheoesophageal fistula (TEF) development caused by pneumatic dilatation (a, b, c) and spontaneous healing of TEF (d).

Lateral pharyngotomy was performed, stenotic cervical esophagus was completely obliterated by several individual stitches, and pharyngostomy was created (Figure 5).

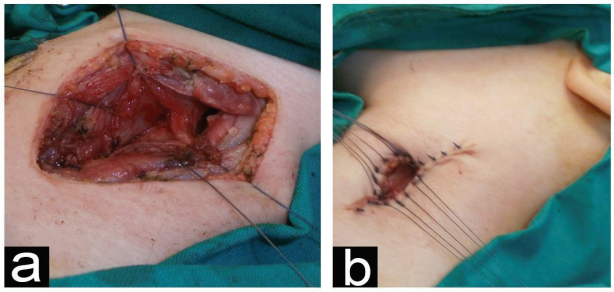

Fig. 5 – Exclusion of the stenotic cervical esophagus (a) and lateral pharyngostomy (b).

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 peristaltic colonic segment, vascularized by left colic vessels (Figure 6).

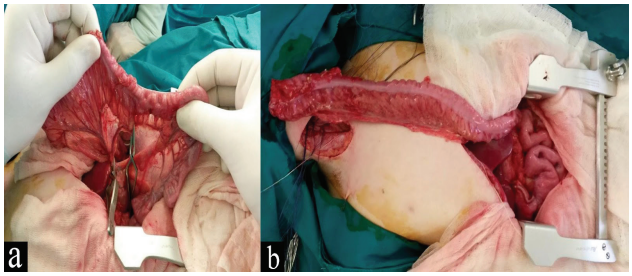

Fig. 6 – Temporary clamping of middle colic vessels and marginal vascular arcade (a) and isoperistaltic colonic conduit vascularized by left colic artery (b).

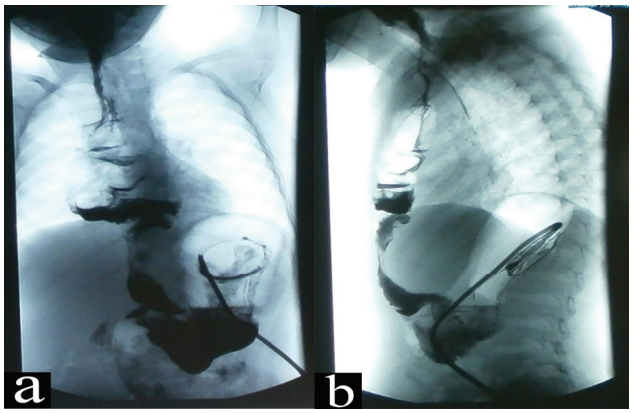

Fig. 7 – Plain (a) and lateral (b) contrast radiographies two weeks after pharyngoesophageal reconstruction with retrosternal colon interposition.

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 [1–4]. According to our best knowledge, only 10 similar cases were reported until now [1–3]. 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 TEF, the presence of pulmonary, mediastinal and pleural infection and signs of sepsis, age, nutritional status and general condition of a patient, together with operative findings are essential for treatment strategy [3, 5, 6].

Symptoms of TEF may be different. The slightest suspicion of TEF requires timely and adequate diagnostic procedure. Esophagography and esophagoscopy can show anatomy of the TEF [7]. Bronchoscopy allows more accurate localization and assessment of TEF morphology [5]. CT scan gives information about inflammatory changes in the mediastinum, pleural cavity and 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 upwards 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 corrosive esophageal stricture 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 TEF, but may help in its spontaneous healing (Figure 4). Lungs protection from aspirated saliva and refluxed gastric content contamination is crucial for 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 TEF, but separates the edges of TEF and does not support the healing [3, 6]. 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 [3, 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 [5].

The staged surgical treatment is an optimal solution for TEF caused by dilatation of a corrosive esophageal stenosis. Our patient had a similar treatment as a young adult with caustic ingestion reported by Crema et al [9]. In our case, the primary repair of TEF was risky. Effective protection of the lungs and the spontaneous healing of TEF was achieved by

exclusion of the esophagus, together with repeated tracheobronchial toilette through an 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

We thank Professor Radoslav Jaković, thoracic surgeon (Faculty of Medicine, University of Belgrade and Institute for Lung Diseases, Thoracic Surgery and Tuberculosis, Clinical Center of Serbia) for his great support in determining therapeutic strategy for our patient.

REFERENCES

1. *Eliçevik M, Alim A, Tekant GT, Sarimurat N, Adaletli I, Kurugoglu S*, et al. Management of esophageal perforation secondary to caustic esophageal injury in children. Surg Today 2008; 38(4): 311–5.
2. *Panieri E, Millar AJ, Rode H, Brown RA, Cywes S*. Iatrogenic esophageal perforation in children: patterns of injury, presentation, management, and outcome. J Pediatr Surg 1996; 31(7): 890–5.
3. *Mutaf O, Avanoglu A, Mevsim A, Özok G*. Management of tracheoesophageal fistula as a complication of esophageal dilatations in caustic esophageal burns. J Pediatr Surg 1995; 30(6): 823–6.
4. *Avanoğlu A, Ergün O, Mutaf O*. Management of instrumental perforation of the esophagus occurring during treatment of corrosive strictures. J Pediatr Surg 1998; 33(9): 1393–5.
5. *Yalçin Ş, Ciftci AO, Karnak I, Tanyel FC, Şenocak ME*. Management of acquired tracheoesophageal fistula with various clinical presentations. J Pediatr Surg 2011; 46(10): 1887–92.
6. *Freire JP, Mendes de Almeida JC*. Review of (acquired) incidental, rare and difficult tracheoesophageal fistula management. World J Surg Proced 2014; 4(1): 9–12.
7. *Ng J, Antao B, Bartram J, Raghavan A, Shawis R*. Diagnostic difficulties in the management of H-type tracheoesophageal fistula. Acta Radiol 2006; 47(8): 801–5.
8. *Imamoğlu M, Cay A, Koşucu P, Ahmetoğlu A, Sarihan H*. Acquired tracheo-esophageal fistulas caused by button battery lodged in the esophagus. Pediatr Surg Int 2004; 20(4): 292–4.
9. *Crema E, Fatureto MC, Gonzaga MN, Pastore R, da Silva AA*. Tracheoesophageal fistula after caustic ingestion. J Bras Pneumol 2007; 33(1): 105–8. (English, Portuguese)
10. *Radovanović N, Simić A, Kotarac M, StojakovD, Sabljak P, Skrobić O*, et al. Colon interposition for pharyngoesophageal postcorrosive strictures. Hepatogastroenterology 2009; 56(89): 139–43.

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