

Case Report

Acquired Tracheoesophageal Fistula After Esophageal Atresia Repair

Boybeyi Türer et al. Acquired Tracheoesophageal Fistula

Özlem Boybeyi Türer, Feridun Cahit Tanyel, Tutku Soyer

Department of Pediatric Surgery, Hacettepe University Faculty of Medicine, Ankara, Turkey

Address for Correspondence: Tutku Soyer, Hacettepe University Faculty of Medicine, Department of Pediatric Surgery, Ankara, Turkey

Phone: +90 532 6651960

E-mail: soyer.tutku@gmail.com

Received: 16 August 2019

Accepted: 19 November 2019

DOI: 10.4274/balkanmedj.galenos.2019.2019.8.60

Cite this article as: Boybeyi Türer Ö, Tanyel FC, Soyer T. Acquired Tracheoesophageal Fistula After Esophageal Atresia Repair. *Balkan Med J*

Abstract

Background: Trachea-esophageal fistula (TEF) recurrence is frequent complication after esophageal atresia (EA) repair. Long new fistulas localized different from congenital TEF sites are called acquired TEFs (acq-TEF). We presented 4 cases to discuss diagnostic and management challenges in different localizations of acq-TEF.

Case report: We retrospectively evaluated the medical records of patients admitted with acq-TEF in last 5 years. From 16 postoperative TEFs, four TEFs were classified as acquired fistulas. Admission age was ranging from 1 to 8 years. Female/Male ratio was 2/2. The presented cases admitted with recurrent respiratory tract infections, choking, coughing. The acq-TEFs were between esophagus and cervical trachea, and right bronchus by passing through intrathoracic abscess cavity, and directly right bronchus, and between colon conduit and trachea. One of acq-TEF was healed spontaneously. Surgical fistula ligation was performed in others.

Conclusion: Acq-TEF is mostly seen secondary to local or diffuse mediastinitis. Besides its classical location of TEF, acq-TEF may be seen at unusual rare localizations such esophagus-to-right bronchus, and conduit-to-trachea. Acq-TEF may be seen at unusual rare localizations that should be kept in mind to evaluate and manage these patients more comprehensively.

Keywords: Children; Congenital Anomaly; Diagnosis; Surgery; Esophageal Atresia; Trachea-Esophageal Fistula

INTRODUCTION

Trachea-esophageal fistula (TEF) recurrence is seen in up to 5% to 10% of esophageal atresia (EA) cases (1). Smithers et al (2) classified TEF as congenital, recurrent and acquired according to the etiology and anatomy. TEF's persists after operation is considered as congenital TEF either missed during initial operation or incomplete repaired ones. Recurrent TEF are fistulas occurred after first successful repair in the same location of the index fistula. Acquired TEF's (acq-TEF) are new pathways between airway and esophagus and occurs different from the original fistula sites (2). They include communications between esophageal anastomosis and pulmonary parenchyma, bronchus or the trachea. Rarely, they can be seen between colon and gastric conduit along with the entire respiratory system. Acq-TEFs have difficult to diagnose and poses a great challenge in the treatment. Therefore, we presented 4 cases to discuss the difficulties in the diagnosis and treatment options of acq-TEF.

CASE REPORTS

The study was performed in adherence to the Declaration of Helsinki and by approval of the Local Ethical Committee. Informed consent has been taken from all cases. We retrospectively evaluated the medical records of patients admitted with postoperative TEF between 2013 and 2019. At total, 42 patients were operated for primary EA and 16 patients were managed for postoperative TEF between 2013 and 2019. Among 16 postoperative TEFs, 11 of them were recurrence, one of them was congenital (missed in the initial operation) and 4 (25%) of them were acquired. The primary repair of 2 cases with recurrent TEF and 1 case with acquired TEF (Case 3) was performed in our center. The primary repair of 13 postoperative TEFs was performed in another center.

The demographic and previous medical histories of the presented cases are given in Table 1. The admission complaints, clinical features and management details are given in Table 2. In all cases, esophagography was

performed and TEFs localized at an unusual localization different from original TEF are diagnosed as acquired TEF. The acq-TEFs were between esophagus and cervical trachea, and right bronchus by passing through intrathoracic abscess cavity, and directly right bronchus, and between colon conduit and trachea (Figure 1).

Case 1 admitted with coughing during feeding and respiratory tract infections lasting for 2 months when she was 3-year-old. Because of poor general condition of the case, we decided to manage the patient conservatively. During this period, oral feeding was ceased and tube feeding and broad-spectrum antibiotics **were recommended**. We observed spontaneous healing of the fistula after 2 months. She has been full oral feeding for the last one year without any respiratory symptoms.

Case 2 admitted with coughing with liquids lasting for 3 months. We detected acq-TEFs between esophagus and cervical trachea and repaired with cervical incision. He has been following uneventfully for the last three years.

Case 3 underwent esophageal replacement with colon conduit supplied by right middle colic artery at 1 year of age. She had leakage from the colo-esophageal anastomosis for three weeks. We detected TEF between cervical trachea and colon conduit. We performed cervical esophagostomy and ligation of fistula between colon conduit and trachea **via cervical incision**. Muscle graft was laid on the sutures. Three months after the operation, she had re-do colon-esophageal anastomosis via median sternotomy. She has been following uneventfully for the last 6 months.

Case 4 admitted with wheezing and coughing lasting for 4 months. The upper GI study revealed TEF to right main bronchus. Right posterolateral thoracotomy was performed to repair the fistula. His postoperative contrast study was normal and he has been full oral feeding for the last 4 years.

DISCUSSION

Postoperative TEF after EA repair is not only diagnostic but also surgical challenge. In most of the large series, all postoperative TEFs were defined as 'recurrent' TEF and there are little known about the acquired fistulas. After classification of postoperative TEFs by Smithers et al (2), acq-TEFs are defined as new communications between esophagus and airway. The most common risk factors in the etiology of TEF recurrence are anastomotic leaks and strictures (3-5). Although balloon dilation is a safe procedure in the treatment of strictures, it is also considered as a risk factor to develop TEF (6). For recurrent TEF, anastomotic leaks cause recanalization of previous pathway. However, acq-TEF occurs as a result of new pathways occurred due to inflammation and mediastinitis. In our patients, three of the cases had prolonged leaks and mediastinitis.

To prove the diagnosis and localization of an acq-TEF can be difficult. A missed congenital TEF should be excluded. These new fistulas can be between esophageal anastomosis and pulmonary parenchyma, bronchus or the trachea, colon or gastric conduit. In diagnosis, contrast esophagography on prone position is reliable method and have positive rate of 94% to detect a fistula (7, 8). Bronchoscopy and catheter insertion through the fistula is also recommended to confirm the diagnosis and to localize the fistula preoperatively. However, we could not insert catheter through the fistula in presented cases because of the abnormal localizations of the fistulas. That is one of the causes which make the diagnosis and surgical management challenging.

The surgical treatment of recurrent and acq-TEFs are more difficult and hazardous than congenital ones because of dense adhesions and mediastinal fibrosis. Therefore, failure rates are common and several fistula repairs may be needed. There is no consensus on the best time and type of surgical repair in recurrent TEFs. The timing of the operation usually depends on the general condition of the patient. Therefore, during prolonged waiting periods, child can be placed on tube feeding. Although there is no consensus, it is usually recommended to wait at least 5 - 6 weeks for resolution of inflammation (3). We reported the spontaneous healing of the acq-TEF during the waiting period in Case 1. To the best of our knowledge, spontaneous healing of an acq-TEF has not been reported so far. Since, acq-TEFs occur after severe inflammation due to leaks, scarring of mediastinum structures may close this new pathway between esophagus and airway. Although it is not possible to make a firm conclusion based on just one case, the possibility of spontaneous healing of fistula may be kept in mind in such cases. In addition, we may only recommend waiting until the general condition of the patient become suitable for operation and inflammation resolves. During the waiting period, tube feeding and antibiotics therapy may be performed. Additionally, a new esophagography may be performed before the surgery if the waiting period is too long.

The surgical access for TEF repair depends on the localization of fistula. Cervical incision can be used for TEFs localized between cervical trachea and esophagus. Thoracotomy is required in TEFs between thoracic esophagus and bronchus or lung parenchyma. Tissue interposition between suture lines is mandatory to avoid recurrence. Pleural, pericardial and muscle flaps are used. In all of our cases we also interposed tissue between suture lines.

In conclusion, acq-TEF is mostly seen secondary to local or diffuse mediastinitis. Acq-TEF may be seen at unusual rare localizations that cause both diagnostic and surgical challenge. The possibility of spontaneous healing may be kept in mind in acq-TEFs.

Figure 1a. The esophagography of Case 1 revealing acquired fistula (arrow) between esophagus and right bronchus passing through the abscess pouch (*).

Figure 1b. The esophagography of Case 2 revealing acquired fistula (arrow) between cervical esophagus and trachea.

Figure 1c. The esophagography of Case 4 revealing acquired fistula between esophagus and right main bronchus (α).

Table 1. Demographic features and previous medical history of the cases with acq-TEF.

Case	Age	Type of atresia	Primary Repair	Bronchoscopy	Trachea-malacia	Anastomotic leak	Number of dilatation	Previous surgical treatments
1	F 4 y	EA + distal TEF	Open technique Another center	No prox fistula	No	Yes	5	-Recurrent TEF repair (x2) - Nissen fund.
2	M 5 y	EA + distal TEF	Open technique Another center	No prox fistula	No	No	3	-Recurrent TEF repair - Nissen fund.
3	F 1 y	Isolated EA	Open technique Our center	No prox fistula	No	Yes	-	Cervical esophagostomy, Esophageal replacement
4	M 8 y	EA + distal TEF	Open technique Another center	No prox fistula	No	Yes	5	Nissen fundo. PSARP

Abbreviations: M: male, F: female, EA: Esophageal atresia, TEF: Tracheoesophageal fistula, Prox: proximal, PSARP: Posterior sagittal anorectoplasty, Fundo: fundoplication

Table 2. The admission complaints, clinical features, and management details of the cases.

Case	Age at admission	Symptom	Duration of symptom	Localization of fistula	Treatment	Prognosis
1	3 y	Coughing RRTI	2 months	Right bronchus (through abscess)	Spontaneous healing	Eventless for 1 year
2	2 y	Wheezing Coughing RRTI	3 months	Cervical trachea	Cervical incision, TEF repair, Muscle flap	Eventless for 3 year
3	1 y	Coughing	3 weeks	Colon conduit to trachea	Cervical incision, TEF repair	Eventless for 6 months
4	4 y	Wheezing Coughing RRTI	4 months	Right main bronchus	Thoracotomy, pleural flap	Eventless for 4 year

Abbreviations: TEF: Tracheoesophageal fistula, RRTI: Recurrent respiratory tract infection