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Treatment of a recurrent tracheoesophageal fistula resulting from prolonged mechanical ventilation. A case review

Leczenie nawrotowej przetoki tchawiczo-przełykowej powstałej w wyniku długotrwałej wentylacji mechanicznej. Opis przypadku

Authors' Contribution:
A – Study Design
B – Data Collection
C – Statistical Analysis
D – Data Interpretation
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F – Literature Search
G – Funds Collection

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ABSTRACT:

Introduction: Tracheoesophageal fistula (TEF) is an abnormal connection between the trachea and esophagus, occurring as either a congenital or acquired condition. Acquired TEF is most commonly associated with malignancy or prolonged mechanical ventilation and may lead to severe respiratory and infectious complications.

Aim: To present a case of recurrent TEF following prolonged intubation and to discuss therapeutic challenges and management strategies.

Case report: A 56-year-old patient with a history of prolonged mechanical ventilation and tracheostomy presented with dysphagia and choking. Endoscopic evaluation revealed a 2.5-cm TEF. Initial surgical management included tracheal resection with end-to-end anastomosis and primary esophageal closure. A small residual fistula persisted postoperatively. Multiple endoscopic interventions, including clipping, suturing, and stenting, were unsuccessful. A second surgical repair also failed, complicated by infection and anastomotic leakage. Ultimately, a third surgical intervention was performed with layered closure and interposition of a pedicled sternocleidomastoid muscle flap. This resulted in complete fistula closure confirmed on follow-up bronchoscopy.

Discussion: Management of TEF includes endoscopic and surgical approaches depending on fistula size and patient condition. Endoscopic methods are typically reserved for small fistulas or high-risk patients. Surgical repair remains the standard for larger or recurrent TEFs. The use of vascularized muscle flaps improves healing and reduces recurrence, particularly in infected or complex cases.

Conclusions: TEF is a life-threatening condition requiring prompt and individualized treatment. In recurrent or large fistulas, incorporation of a muscle flap significantly enhances treatment success and reduces the risk of recurrence.

KEYWORDS: endoscopic treatment, surgical treatment, tracheoesophageal fistula

STRESZCZENIE:

Wstęp: Przetoka tchawiczo-przełykowa (TEF) jest nieprawidłowym połączeniem między tchawicą a przełykiem. Może ona występować jako wada wrodzona lub schorzenie nabyte. Nabyta TEF najczęściej związana jest z chorobą nowotworową lub przedłużoną wentylacją mechaniczną i może prowadzić do ciężkich powikłań oddechowych oraz infekcyjnych.

Cel: Przedstawienie przypadku nawrotowej TEF po długotrwałej intubacji oraz omówienie trudności terapeutycznych i strategii leczenia.

Opis przypadku: Pięćdziesięcioześcioletni pacjent z wywiadem długotrwałej wentylacji mechanicznej i tracheostomii zgłosił się z zaburzeniami połykania i krztuszeniem. W badaniu endoskopowym stwierdzono przetokę tchawiczo-przełykową o długości 2,5 cm. Początkowe leczenie chirurgiczne obejmowało resekcję tchawicy z zespoleniem koniec do końca oraz pierwotne zamknięcie przełyku. Po operacji utrzymywała się niewielka przetoka resztkowa. Liczne interwencje endoskopowe, w tym klipsowanie, szycie oraz stentowanie, okazały się nieskuteczne. Druga operacja również zakończyła się niepowodzeniem, powikłanym zakażeniem i nieszczelnością zespolenia. Ostatecznie wykonano trzeci zabieg chirurgiczny

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z warstwowym zamknięciem przetoki oraz użyciem uszypułowanego płata mięśnia mostkowo-obojczykowo-sutkowego. Uzyskano całkowite zamknięcie przetoki, potwierdzone w kontrolnej bronchoskopii.

Dyskusja: Leczenie TEF obejmuje metody endoskopowe i chirurgiczne, w zależności od wielkości przetoki i stanu pacjenta. Techniki endoskopowe stosuje się głównie w małych przetokach lub u pacjentów wysokiego ryzyka. Leczenie chirurgiczne pozostaje standardem w przypadku dużych lub nawrotowych przetok. Zastosowanie unaczynionych płatów mięśniowych poprawia gojenie i zmniejsza ryzyko nawrotu, szczególnie w przypadkach powikłanych zakażeniem lub o złożonym przebiegu.

Wnioski: TEF jest stanem zagrożenia życia wymagającym szybkiego i zindywidualizowanego leczenia. W przypadku dużych lub nawrotowych przetok zastosowanie płata mięśniowego istotnie zwiększa skuteczność terapii i zmniejsza ryzyko nawrotu.

SŁOWA KLUCZOWE: leczenie chirurgiczne, leczenie endoskopowe, przetoka tchawiczo-przełykowa

ABBREVIATIONS

CT – computed tomography
TEF – tracheoesophageal fistula

INTRODUCTION

Tracheoesophageal fistula (TEF) is defined as an abnormal communication between the tracheal and esophageal lumens. It can be either a congenital defect or an acquired condition. Congenital fistulas are rare and occur in roughly 1 in 2500 live births [1]. The most common cause of the acquired tracheoesophageal fistula is malignancy, which accounts for around 50% of cases [2]. Non-malignant TEF is caused in up to 75% by prolonged mechanical ventilation [3], other causes are granulomatous mediastinal infections, neck trauma, and surgeries on the esophagus or the trachea [4–7]. Low-pressure endotracheal tube cuffs reduce the incidence of this complication; nevertheless, it complicates up to 3% of long-term intubations [8]. Fistulas can develop after an average of 40 days of the mechanical ventilation, but the individual cases can range from 12 to 200 days [9]. Patients with TEF present with cough, aspiration, fever, dysphagia, chest pain, recurrent pneumonia, serious respiratory failure, or even cardiopulmonary arrest [10]. TEF can be a life-threatening condition, and if untreated, it can lead to death as a result of severe infections [11].

AIM

The aim of this report is to present a case of a recurrent tracheo-esophageal fistula following prolonged intubation.

CASE REPORT

A 56-year-old patient was admitted to the department of thoracic surgery with dysphagia and choking during meals. Symptoms had been present for two years, following prolonged mechanical ventilation with established tracheostomy following an acute post-traumatic subdural hematoma.

In the process of diagnosis, gastroscopy and bronchoscopy were performed. They visualized a fistula roughly 2.5 cm in diameter, located 3 cm inferiorly from the lower border of the larynx and 22 cm from the incisor line. Trachea under the fistula showed no abnormalities. The patient was qualified for surgery in order to close TEF.

A transverse cervical incision over the sternum was performed, the underlying tissues were carefully dissected, and the upper trachea was exposed. The visualized trachea was covered by fibrous adhesions, and a tracheostomy scar was noted. After releasing the adhesions and transecting the fistula, a 1.5-cm segment of trachea was resected. The esophageal lesion of about 3 cm was visualized and closed directly using a double-layer running suture. The trachea was anastomosed end-to-end using a running suture; tissue glue was used to seal the anastomosis. Thorough hemostasis was achieved, and layered closure of the wound was performed. Histopathological analysis of the resected fragment of the trachea showed fibrosis of the mucous membrane and focal ossifications of cartilage.

After surgery, total parenteral nutrition was administered. In the control gastroscopy, 5 days after surgery, a residual fistula measuring about 1–2 mm was observed. Through the opening, a mucous content was aspirated and expelled during respiratory movements.

A gastroscopy was performed, and an attempt to close the residual opening in the esophagus using a hemostatic clip was made. Due to fibrosis around the fistula and tissue stiffening, the procedure was unsuccessful. In the following days, a second gastroscopy, this time under general anesthesia, was performed. An attempt to close the fistula using the SutuArt suturing set was made but the first attempt was unsuccessful, and due to significant upper esophageal sphincter edema further attempts were abandoned. In the third gastroscopy, a remaining 3–4-mm opening in the esophagus wall was shown, a Hanarostent through-the-scope esophageal stent was placed in the esophagus under X-ray control. Control radiograph after a week confirmed a stent placement in the esophagus. Three weeks after the stent deployment, a control gastroscopy was performed, and the stent was removed. After removing the stent, a TEF was noted, its diameter measuring about 1 cm. The fistula was temporarily closed using the X-tack endoscopic suturing system.

To surgically close TEF, the patient was transferred to the otorhinolaryngology ward. A surgical repair was performed. An incision on the right side of the neck was made. Soft tissues of the neck were systematically and meticulously dissected. The lobe of the thyroid gland, side of the pharynx, trachea, esophagus, and main blood vessels of the neck were visualized. The trachea was dissected from the esophagus, and the fistula was localized. Mucus

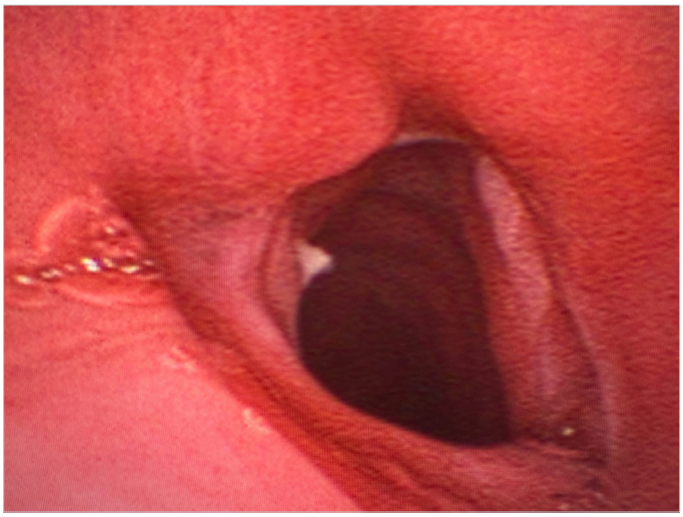


Fig. 1. Bronchoscopic view of a fully closed fistula after treatment with a partially absorbed suture material.

and muscle layers of both trachea and esophagus were dissected. A layered closure of both structures was performed. Single sutures were used on the mucosa, and running sutures were used on the muscular layer. A hemostasis of the surgical site was performed, and the skin was closed in layers. Following the surgery, a nasogastric tube was placed in the patient's stomach to feed them.

In the control neck and chest CT, gas in all compartments of the neck was described, indicating anastomotic leakage. Eight days after surgery, the C-reactive protein level reached 12.8 mg/l, and the wound dressing was soaked with blood-tinged serous discharge. A wound swab was taken for microbiological examination, and an empirical antibiotic therapy was initiated. Wound cultures grew *Pseudomonas aeruginosa*, *Klebsiella pneumoniae* ESBL+ and MRSA; antibiotic therapy was adjusted. Twenty days after the second surgery, the patient started drinking fluids, and the following day, the nasogastric tube was removed. No pulmonary symptoms were noted, and in a control radiogram, subcutaneous emphysema decreased. A control gastroscopy was conducted and showed a fistula big enough for a narrow gastroscope to pass through the opening. A nasogastric tube was inserted again; the patient started enteral feeding.

A third surgical intervention was performed. The surgical technique was nearly the same as in the previous one. Additionally, to accommodate healing of the fistula, the pediculated flap from the sternocleidomastoid muscle was moved and secured between the esophagus and trachea after directly closing these structures with sutures.

Control radiogram showed no emphysema. Seven days after the third surgery, the patient reported abdominal pain and diarrhea. The surgical dressing was soaked with yellowish discharge. A wound swab was taken. A CT scan of the abdomen was performed, and signs of inflammation in the rectum and sigmoid colon were described. *Clostridioides difficile* antigen test from stool was negative. The swab results revealed the presence of the same bacteria as in the previous one. A clinical pharmacology consultation was requested, and the antibiotic therapy was adjusted according to the recommendations. Cultures from blood and the central line were obtained for microbiological analysis, but only contamination with skin flora was noted. Twenty days after the third surgery, a gastroscopy was performed; no fistula was visualized. The patient was discharged with recommendations for outpatient follow-up. During a control bronchoscopy, a closed fistula was visualized with a partially absorbed suture material (Fig. 1.).

DISCUSSION

Management of TEF includes conservative, endoscopic, and surgical techniques. Conservative therapy is rarely used and limited only to specific cases [12].

During gastroscopy, various procedures can be used, the most common ones being the application of fibrin glue, ablation, placing sutures or clips, and tracheal or esophageal stents [13]. Endoscopic procedures are commonly applied for small fistulas, patients who are not suitable for an operation, or as a palliative therapy in malignant fistulas [14].

Various surgical techniques are employed in the repair of TEF, with neither being universally superior, and the choice should be made based on the size and location of the fistula and the condition of the trachea [11].

Direct fistula repair is usually used in fistulas measuring under 1 cm in diameter. Anterior and lateral approach can be used [15]. The fistula is transected, and both trachea and esophagus are closed in layers. Strap muscles such as the omohyoid are then mobilized and secured over the esophageal repair, which lowers the probability of recurrence [16].

In bigger fistulas or if primary closure is not possible, resection of the trachea is performed. The resected fragment should not exceed 4 cm to prevent excess tension [17]. An end-to-end tracheal anastomosis is then performed. The esophagus is directly closed, and a strap muscle is placed over it to reinforce the repair [18].

In the surgical treatment of large and complex TEF that cannot be anastomosed directly after fistula resection, autologous tissue flaps can be used. However, it is not a routine treatment. A pedicled sternocleidomastoid musculocutaneous flap was reported to repair a large TEF with the cutaneous component serving as the new membranous wall [19]. In a retrospective study, the application of extrathoracic muscle flaps was explored; the most common choice was latissimus dorsi, followed by pectoralis major and serratus anterior. Flaps were used to repair the esophagus, trachea, or, in some cases, both structures [20]. In a series of 8 cases, the authors describe a perforator-based intercostal artery muscle flap used in different types of intrathoracic fistulas [21]. Some authors described a bipaddled radial forearm free flap in TEF reconstruction [22, 23]. Other described flaps are the gastric antral patch, posterior pedicled pericardial flap, and autologous dermal flap reinforced with a pedicled pectoralis major muscle and rib cartilage graft [24–26].

Other methods of TEF management are the repair of the membranous wall using synthetic patches and bioprosthetic

materials or esophageal diversion, either as esophageal exclusion or the "double patch" technique. They are mainly employed if other procedures are not effective [27–30].

In our case, the primary surgery comprised tracheal segment resection and direct esophagus closure; however, a muscular flap was not used. Following the procedure, the fistula recurred, and many repair attempts using endoscopic techniques were unsuccessful. The second surgery turned out to be unsuccessful and only the third one, where a muscular flap was used, resulted in full recovery. That highlights the effectiveness of muscular flaps in accelerating healing and preventing recurrence of TEF. Autologous flaps prove to be effective, especially when the surgical site is infected.

CONCLUSIONS

TEF can be a life-threatening condition and should be treated as soon as possible. Different endoscopic or surgical methods can be employed, but the choice should be made based on the site and size of the lesion. In larger TEFs a muscular flap should be used to ensure better healing and lower the chance of recurrence.

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Table of content: https://otorhinolaryngologypl.com/issue/17179 Tables: – Figures: 1 References: 30

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