ENDOSCOPIC TREATMENT OF ESOPHAGEAL-BRONCHIAL FISTULA IN A PATIENT WITH A LEYDEN MUTATION


Annotation. Clinicевский случай успешного эндоскопического хирургического лечения пищеводно-бронхиального свища у пациента с Мутацией Лейдена и хронической тромбоэмболией легочной артерии.

Abstract. Bronchoesophageal Fistula (BEF) is an uncommon condition related to complex thoracic surgery. The development of BEF is usually an indication of the progression of bronchial or lung cancer; whereas the etiology of this illness being recorded as benign is much rarer – not exceeding 4 - 6% [1, 2].

Surgery is the main method for treating patients who have benign BEF, allowing for the reliable result. At the same time, similar operations are extremely traumatic and are fraught with the development of post-operation complications, especially when concerning patients with genetically determined coagulopathy.

Factor V Leiden mutation is a hereditary coagulopathy in which there is a point mutation in the gene that encodes blood coagulation factor V. Factor V Leiden mutation is the most common cause of the hereditary disposition to thrombosis, heart attacks, strokes in Europeans. The frequency of occurrence among the population of the USA is 4 – 6% [3].

We present our own observation of successful endoscopic surgery to treat BEF in a patient with Factor V Leiden mutation and chronic pulmonary embolism.

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ENDOSCOPIC TREATMENT OF ESOPHAGEAL-BRONCHIAL FISTULA IN A PATIENT WITH A LEYDEN MUTATION

Sklifosovsky Research Institute for Emergency Medicine

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ENDOSKOPIČESKOE LÉČENIE PİŞČEVDÔNO-BRONHÌAL'NOGO SVIÇHA 
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Bronchoesophageal fistula, fistula, bronchus, esophagus.

Key words: endoscopy, bronchoscopy, esophagoscopy, Factor V Leiden, Bronchoesophageal fistula, fistula, bronchus, esophagus.

Aim of the paper
To share our own experience of endoscopic surgery using the tunnel technique to treat BEF in patients who have hereditary coagulation.

Main body
Patient K., 58 years old, was transferred to the Sklifosovsky Research Institute for Emergency Medicine from another facility on 04.12.2017 for treatment for a BEF.

From the patient’s medical history it is know that they have suffered from hypertension for a long time, acute cerebral accident with left-sided hemiparesis and pulmonary embolism (PE). The diagnosis for hereditary coagulation was then verified as Factor V Leiden. Long-term mechanical lung ventilation was performed, which required the application of a tracheostomy. 16 days after tracheal decannulation, BEF was detected during an endoscopic investigation. A percutaneous endoscopic gastronomy (PEG) was performed in order to allow for enteral feeding and to prevent aspiration.

Upon admission to the Sklifosovsky Research Institute for Emergency Medicine, the patient complained of having a persistent cough with phlegm, and feeling weak and out of breath. The patient was fed by PEG exclusively and saliva was spat out.

Chest x-ray: local pneumosclerosis located on the right in the middle region of the lung field, no abnormal content found in the pleural cavity.

Chest CT Scan: fistula was not visualised. Infiltrative changes in the right lung with multiple cavity lesions formation were present.

Scintigraphy determined signs of a 2-sided polysegmental PE with a perfusion deficit of 35-40%.

Bronchoscopy: 10–15 mm caudal bifurcation of the trachea along the posterior wall of the left main bronchus, an 8x3 mm sized oval-shaped deformity (fistula opening) with an inflow of purulent content from its lumen was detected.

Esophagoscopy: At a length of 25 cm from the incision, a deformation of the oesophagus due to displacement from the outside along the right side of the anterior wall was detected, on which a 6x2 mm sized, slit shaped fistula opening with epithelized edges was seen. When inserted the dye (methylene blue) was observed to have entered into the lumen of the left main bronchus.

Coagulation indicators at the moment of hospitalisation (taking into account heparin injection of 20,000 units per 24 hours): Prothrombin 62.7%, INR 1.45, aPPT 27.5 secs.

Coagulation indicators at the moment of hospitalisation: Prothrombin 66%, INR 1.24, Fibrinogen 4.99 g/L, aPPT 25.3 secs.

A year after the stent was inserted (December 2018), the patient was admitted again to the Sklifosovsky Research Institute for Emergency Medicine on a routine basis.

During an X-ray contrast examination of the oesophagus, contrast agent leakage was noted in the left main bronchus through the fistula passage at the T4 level, which indicated that the functional spinal unit was preserved and that there was no effect from the stentography.

Bronchoscopy confirmed the satisfactory position of the bronchial stent. When performing esophagoscopy, some reduction to the size of the fistula (3 mm in diameter) with epithelized edges was determined.

No comparison with the CT and scintigraphy data from 2017 was made.

Coagulation indicators (taking into account an intake of 20 mg of Rivaroxaban per 24 hours): Prothrombin 63.8%, INR 1.47, aPPT 25.3 secs.

The patient was discharged with the recommendation to have further dynamic observation and for food intake to be via PEG.

Check-up a year later (December 2019) - the patient had the same complaint of having a cough and choking when accidentally swallowing saliva. A gradual reduction in body mass index was noted (weight 43 kg, height 154 cm, BMI 18.13).

Coagulation indicators (taking into account an intake of 20 mg of Rivaroxaban per 24 hours): Prothrombin 66%, INR 1.24, Fibrinogen 4.99 g/L, aPPT 25.7.

Endoscopic examination confirmed that the functional spinal unit was present despite the fistula area being protected by a stent for two years. Taking into consideration the high risk of surgical intervention for the patient, minimally invasive endoscopic treatment was proposed; the purpose of which being removal of the bronchial stent and removal of the fistula using the tunnel technique in the submucosa layer of the oesophagus.
Operation on 17.12.2019: Under complete intravenous anesthesia with high-frequency mechanical lung ventilation, intubation of the trachea was performed with a 10 mm rigid bronchoscope tube. With the help of rigid forceps, the stent was removed from the lumen of the left main bronchus.

Subsequently, the rigid tube was then changed for an orotracheal intubation tube with volumous ventilation. When inspecting through the orotracheal tube, along the posterior wall of the left main bronchus granulation tissue was found without significant narrowing of the lumen.

In the following step, the gastroscope was inserted into the lumen of the oesophagus. The mouth of the oesophagus was 15 cm from the incisions. The oesophagus was easy to pass through and wasn't malformed. A fistula hole with epithelized edges of 2 mm in diameter was identified along the anterior oesophageal wall at 12 cm distal to the pharyngeal-oesophageal junction. The lower oesophageal sphincter (LES) is found 41 cm from the incisions.

With the help of an injection needle, at 5 cm proximity to the functional spinal unit along the anterior oesophageal wall a sterile saline solution tinted with an indigo carmine solution (10 ml) was injected. A thick submucosal infiltrate formed. Along the proximal edge of the infiltrate, a longitudinal dissection of the mucus membranes of the oesophagus was performed with an Triangle Tip Knife 1.5 cm in length. Next, using the cap on the distal end of the endoscope and the dissection knife in SPRAY COAG mode, a submucosal canal, or "tunnel" 4 cm in length and 1.5 cm in width was formed. In the area of the functional spinal unit, the submucosal layer consisted of fibrous tissue.

Using the dissection knife, the submucosal layer was mobilised around the fistula with an excision of the fistula passage at the bronchial wall, with coagulation of the bronchial canal of the fistula. The fistula passage was twisted into the oesophageal lumen in the form of a sleeve; the edges of the malformation were subsequently clipped to the membranes of the oesophagus at the level of the entry to the tunnel and the area of the twisted fistula passage with clips. Upon inspection there were no signs of bleeding, and the edges of the malformation were brought together tightly.

Histological examination of the excised fistula: fragments of granulated skin and mucus membranes of the oesophagus and bronchus with signs of chronic inflammation.

The patient was discharged from the ward in a satisfactory condition on the 13th day after the operation.

During a check-up a month later, the patient didn’t have any complaints. During an X-ray contrast examination of the oesophagus with barium sulphate suspension, no leakage beyond the contours of the oesophagus was detected. During a bronchoscopy on the posterior wall of the left main bronchus, a longitudinal scar was found without malformation and narrowing of the lumen. Oesophagoscopy in the area which was clipped found 2 longitudinal scarring with sporadic granulation up to 2 mm in diameter. The patient began to eat by mouth. A month later the PEG was removed.

Discussion:
Sporadic observations of effective stenting of the oesophagus in the area of disjointed fistula have been published [4]. In our observation, stenting the left main bronchus over a period of two years didn’t lead to obliteration of BEF. At the same time, the long-term presence of the endobronchial covered stent caused granulated skin to grow on its cranial and caudal ends.

In recent years, reports are increasingly appearing regarding the successful use of the endoprosthetic method in patients with BEF benign in nature [1].

A team of authors [5] compiled the results of 9 years of experience in diagnosing and treating adult patients with benign BEF. The analysis of this data has shown that the initial endoscopic intervention when treating BEF wasn’t effective, and repeat intervention in different forms (clipping, stenting, applying histoacryl tissue glue) proved effective in 29% of cases observed.

In the clinical observation that we have presented, the patient with BEF of an uncertain etiology with a background of genetically determined coagulation and recurrent PE, the minimally-invasive endoscopic method with creating a tunnel in the submucosal layer of the oesophagus allowed for the mobilisation of the fistula canal around its entire circumference, and to excise it from the base (the walls of the left main bronchus); minimising the risks of complications developing.

Conclusion: The endoscopic excision of the oesophageal-bronchial fistula using the tunnel method with airtight clipping of the defect of oesophageal mucous membranes may be an effective and safe treatment method for patients with this condition.
Img. 1: Bronchoesophageal Fistula

Img. 2: Self-expandable silicone stent in the left main bronchus

Img. 3: BEF on X-Ray in a year after stenting
**Img. 4: Plan of operation**

**Img. 5: Histologic images of BEF**
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ДИАГНОСТИКА МАТЕМАТИЧЕСКОГО ДОКАЗАТЕЛЬСТВА ГИПОТЕЗЫ БИЛЯ В МЕДИЦИНСКОЙ ПСИХОЛОГИИ (РЕМЕЙК ПРЕДЫДУЩИХ СТАТЕЙ АВТОРА О ВЕЛИКОЙ ТЕОРЕМЕ ФЕРМА)

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DIAGNOSTICS OF MATHEMATICAL PROOF OF THE BEAL CONJECTURE IN MEDICAL PSYCHOLOGY (REMAKE OF PREVIOUS AUTHOR’S ARTICLES CONCERNING FERMAT’S LAST THEOREM)