

Esotracheal Fistula in Esophageal Stenoses of Malignant Origin - Case Report

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Rezumat

Fistula esotraheală în stenozele esofagiene maligne - prezentare de caz

Introducere: Aproximativ 80% din fistulele esotraheale și mediastinale dobândite sunt de origine malignă. Apariția fistulei reprezintă o complicație devastatoare atât pentru pacient cât și pentru medic, iar în absența tratamentului supraviețuirea este între 1 și 6 săptămâni.

Material și metodă: prezentăm cazul unui pacient de 51 de ani, fumător, cu disfagie progresivă, inițial pentru solude, apoi pentru semisolide, ulterior pentru lichide, urmată de tuse la fiecare deglutiție, secundară dezvoltării unei fistule esotraheale și incapacitatea de alimentație ceea ce a condus la declinul stării generale. Intervenția chirurgicală a constat în montarea unei endoproteze esofagiene prin foraj transtumoral care a asigurat și ocluzia traiectului fistulos.

Rezultate: Evoluția postoperatorie a fost favorabilă, alimentație per os se reia la 8 ore postoperator cu toleranță digestivă bună. Examenul radiologic cu substanță de contrast, efectuat la 48 de ore postoperator evidențiază proteza endoesofagiană permeabilă, fără reflux al substanței de contrast în arborele traheobronșic.

Concluzii: Montajul laparogastroscopic prin foraj transtumoral utilizând proteze semirigide siliconate reprezintă singurul tratament paliativ eficient în cazul fistulelor esotraheale sau esobronșice.

Cuvinte cheie: malignitate, fistulă esotraheală, montaj laparogastroscopic

Abstract

Background: Approximately 80% of acquired eso-tracheal or mediastinal fistulae are of malignant nature. The occurrence of an eso-respiratory malignant fistula is a devastating complication for both patient and doctor, and, if not treated, records a survival time of 1 to 6 weeks.

Material and method: We present a patient, aged 51, smoker, with progressive dysphagia, at first to solids, then to semi-solids and liquids, followed by manifestation of post-deglutition cough due to eso-tracheal fistula, incapability of feeding, and decline of the general condition. Surgical intervention consists of esophageal transstenotic endo-prosthesing by transtumoral drilling with prosthetic montage and the occlusion of the eso-tracheal fistulous orifice.

Results: The post-operative evolution is favorable, the feeding per os resumes in 8 hours after surgery, good digestive tolerance. The radiologic examination using contrast medium, performed in 48 hours after surgery, reveals a permeable esophageal endo-prosthesis, without reflux of contrast substance in the tracheobronchial tree.

Conclusions: The laparogastroscopic montage of prosthesis

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through transtumoral drilling, using siliconized semi-rigid prostheses, represents the only efficient palliative treatment of malignant eso-tracheal or eso-bronchial fistula.

Key words: malignancy, eso-tracheal fistula, laparo-gastroscopic montage

Introduction

Esophageal neoplasm is currently the seventh cause of death from cancer in the world. The disease's incidence varies widely. In the USA, it ranges between 3 and 4 per 100.000 people, with a decreasing tendency during the last 20 years. In Europe, this incidence ranges from 1.8 per 100.000 people in Finland to 13.6 per 100.000 people in France, while in the other European countries its level is of approximately 2-3 cases per 100.000 people. The highest incidence however is to be found in countries such as Iran and Afghanistan, and in the Northern part of China, with levels going up to 144 cases per 100.000 people.

From among developing countries in Europe, Romania ranks fifth as regards esophageal cancer (5,4‰) and has one of the most unfavorable prognosis for cancer. Due to the specific intra-thoracic topography, clinically inaccessible and oligo-symptomatic, the disease is rather incurable when dysphagia occurs, the first conclusive sign for the patient – as claimed by theory and the clinician, a sign rather ignored however, for a long period of time, by the afflicted person.

Less than 50% of the patients with dysphagia secondary to esophageal cancer are given a radical treatment, due to the locoregionally advanced stages and the distant disseminations. Cases of one-year survival represent less than 20%, while 10 to 15% of the patients cannot be exposed to the risk of a surgical intervention, palliative or not, resectability without other expectations being qualified as adventurous.

Approximately 80% of acquired eso-tracheal or mediastinal fistulae are of malignant nature (1). As noticed by Burt and collaborators (2), most of them (77%) are caused by esophageal neoplasm, followed by pulmonary neoplasm (16%), by tracheal, laryngeal and thyroidal neoplasm, as well as, to a lesser extent, by metastatic mediastinal adenopathies (2%). The occurrence of an eso-respiratory malignant fistula is a devastating complication for both patient and doctor, and, if not treated, records a survival time of 1 to 6 weeks (2).

Mechanic ventilation is the most frequent cause of benign eso-tracheal fistulae; other less frequent causes include iatrogenic lesions, foreign bodies, traumatism, antecedents of surgical interventions on the trachea or esophagus, stents, mediastinal granulomatous infections (tuberculosis, histoplasmosis), AIDS (3, 4).

Benign eso-tracheal fistulae receive initially a conservative treatment, followed by a definitive surgical intervention that may vary, depending on the dimension and location of the lesion, from fistula removal to the resection and reconstruction

of the trachea.

For patients with malignant eso-tracheal fistulae, the curative treatment depends - according to the majority of authors - on the surgical resectability of the primary tumour and the trajectory of the fistula, most of the patients presenting the disease in advanced stages, when only palliative treatments can be performed.

In these situations, the therapeutic objectives aim at giving the patient the possibility to feed himself (an objective easily achievable, in a mini-invasive way, by means of gastrostomy or jenuostomy, procedures which are nevertheless extremely invalidating, transform the patient into a psychologically and socially disabled person and, indirectly, speed up the exitus), hence ensuring a satisfactory nutritional status allowing for the beginning of oncologic therapy (chemotherapy and radiotherapy) and for a limited protection of the respiratory tree, in cases in which eso-tracheal fistula occurs, by allowing oral evacuation of salivary secretions, and ingestion and deglutition, in general.

As one of the non-invasive palliative interventions, the endoscopic montage of a prosthesis, rarely achievable and almost never facile, meets these requirements in general, with the exception of the price (gastroenterologists prefer flexible prostheses, metallic or not, which are expensive), and of the percentage of prothesing failures (25-45% for esophageal tumors and significantly increased for eso-tracheal fistulae). These failures are caused by: the technical impossibility of crossing the tumoral stenosis with the endoscope; the endoscopist's reluctance to approach "sensitive areas", such as the esophagus poles (extremities); the presence of an eso-tracheal or eso-bronchial fistula of large caliber, which causes a brutal imbalance during endoscopic examination, aggravated by the considerable dimensions of a fistula that cannot be effectively occluded with a flexometallic prosthesis, permeable or not; the major problems arising from standard endoscopic examination of the esophagus with air insufflation and serum lavage, due to the fistula and the invasion of the respiratory tract with air, which sometimes leads to respiratory arrest.

The montage of the prosthesis in open surgery applies to cases in which laparo-gastroscopic prothesing is not possible (tumor infiltrated cardia, with the axis significantly displaced, non-discernible, tough tumors requiring digital drilling etc.) and usually, given the location, besides tracheobronchial comorbidities, it faces the risk of open surgery complications, a risk that is much higher for patients with advanced esophageal neoplasms, cachectic, anemic and presenting severe hypoproteinemias (eviscerations, suppurations, eventrations, etc.).

The laparo-gastroscopic prothesing under general anaesthesia with orotracheal intubation, with the adjusted insertion of the tracheal tube (an original option) may occlude or go beyond the fistulous orifice, with or without selective intubation of one of the main bronchi. The approach and the method are original options; they use semi-elastic prostheses made of plastic material, in a wide range of forms, dimensions and shapes, with a view to individualising the therapy from a technical point of view too.

Case report

Under these circumstances, the only effective and viable solution is the insertion of a prosthesis, not endoscopically, but surgically, using the mini-invasive option, not by pushing a flexometallic prosthesis, in a relatively elastic and indirect way, but by performing transtumoral traction and drilling with a semi-rigid siliconized prosthesis, a procedure unique in the world in terms of conception, method and fabrication, which was awarded the First Prize in Japan, in 2005, at the Yokohama International Gastric Cancer Congress.

This procedure has multiple advantages. It enables the oral, normal feeding and the improvement of the psychic condition of patients, who are seriously afflicted by the disease, both physically and psychically (this is probably the best perceived advantage); it offers a solution in cases where endoscopic prosthetic montages are not feasible (otherwise candidates to gastro or jejunostomy); the prosthesis occludes the fistulous aerodigestive, digestive-mediastinal or pleural tract; it speeds up the post-operative healing (absence of abdominal incision); it allows for a rapid beginning or resuming of oncologic therapy.

One of these cases is patient CC, aged 51, smoker of 20 cigarettes per day during 30 years, admitted to hospital for dysphagia, coughing spells, post-deglutition cough, and significant weight loss of about 8 kg in 2 months.

The current disease has occurred approximately 3 months ago, insidiously, with progressive dysphagia, at first to solids, then to semi-solids and liquids, followed by manifestation of post-deglutition cough, incapability of feeding, and decline of the general condition.

The objective examination reveals that the patient has a satisfactory general condition, is underweight, IMC, with sallow teguments and fetid breath.

The radiologic examination using contrast medium reveals a slower deglutition, fractioned, symmetric pyriform valleculae and sinuses; eso-tracheal fistula at the carina level with minimum right bronchial passage (at the first swallowing of contrast substance) (Fig. 1); administration of contrast substance is interrupted.

The upper digestive endoscopy reveals that the esophageal lumen, at approximately 35 cm from the dental arcade, is narrowed by a vegetant tumor; biopsy is performed. The stenosis is surpassed with the endoscope and a guiding catheter is inserted. Cardia, stomach, duodenal bulb, D1 - normal. The histopathology result: pulmonary adenocarcinoma poorly differentiated.

Surgical intervention is performed under general anaesthesia with orotracheal intubation; it consists of: exploratory laparoscopy for abdominal evaluation; laparo-gastroscopy with lower gastric and esophageal visualisation, and esophageal transthenotic endo-prosthesing by transtumoral drilling with prosthetic montage performed by traction and the occlusion of the eso-tracheal fistulous orifice, followed by minimal gastrorrhaphy and peritoneal drainage.

The post-operative evolution is favorable, the feeding per



Figure 1. Malignant esophageal stenosis with eso-tracheal fistula at the carina level with minimum right bronchial passage (at the first swallowing of contrast substance)

os resumes in 8 hours after surgery, good digestive tolerance, precocious deglutition without alterations.

The radiologic examination using contrast medium, performed in 48 hours after surgery, reveals a permeable esophageal endo-prosthesis, without reflux of contrast substance in the tracheobronchial tree – occluded fistula (Fig. 2).

The patient returns after 7 days from hospital discharge, with food stuck in the prosthesis, as result of feeding disorders due to excessive trust. The endoscopic examination – because of air insufflation and pneumatization of the occluded esophagus – triggers off the invasion of the tracheobronchial tree with air, which produces a respiratory arrest rapidly resuscitable. Once the endoscopic examination interrupted, it is decided to revisit the montage by laparo-gastroscopic means; it is noticed that the montage is stable and the prosthesis is permeable.

The post-operative evolution was simple, with systematic clinical and radiologic examinations, given the novelty of the method (Fig. 3).



Figure 2. Permeable esophageal endo-prosthesis, without reflux of contrast substance in the tracheobronchial tree

Discussions

We believe that the current standard in palliative treatment of advanced esophageal neoplasm – once a candidate for gastrostomy or jejunostomy – is the endoscopic montage of self-expandable metallic endoluminal prostheses, with impermeable walls or, rather, the laparo-gastroscopic montage of semi-rigid endoluminal prostheses, siliconized or not, made of plastic material (3, 5).

Unfortunately, most frequently the endoscopic montage of prostheses is not possible in the case of eso-tracheal or eso-bronchial fistulae, due to air insufflation that accompany endoscopy, which triggers severe coughing spells and may result in respiratory arrest. Another shortcoming of endoscopy resides in the type of prosthesis – the flexometallic one, the only one that can be inserted endoscopically, which presents difficulties of insertion, due to the relatively blind montage.

If we take also into account, in addition to these disadvantages, the high price of the flexometallic prosthesis, increased by the need to ensure impermeability of the prosthetic wall, while the beneficiary is an incurable patient



Figure 3. The patient after the second intervention

with a low life expectancy, pauper in most cases, we believe that turning to surgical treatment is well justified.

Conclusions

In this context, the laparo-gastroscopic montage of prosthesis through transtumoral drilling, using siliconized semi-rigid prostheses, represents the only efficient palliative treatment of malignant eso-tracheal or eso-bronchial fistula.

Historically, eso-tracheal fistulae of malignant origin have been considered practically irreducible, and have been directed, without real perspectives unfortunately, to thoracic or general surgery of the upper digestive tube. In post-caustic benign fistulae, the cvasi-complete esophagectomy with esophageal muscular layer remnant postero-tracheal and its suture have represented the surgical solution and the subject of a PhD thesis.

Post-laryngectomy eso-tracheal fistulae represent a special subject and a separate chapter, because of radiotherapy, possibly cancer recurrence; hence, they should be examined through a differentiated approach.

References

1. Singhal SK, Virk RS, Dass A, Sandhu BS. - Tracheoesophageal fistula: New option in management.

- Indian J Otolaryngol Head Neck Surg. 2006 Jul;58(3):300-2.
2. Burt M, Diehl W, Martini N, Bains MS, Ginsberg RJ, McCormack PM, Rusch VW. - Malignant esophagorespiratory fistula: management options and survival. *Ann Thorac Surg.* 1991 Dec;52(6):1222-8; discussion 1228-9.
 3. Stemerma DH, Caroline DF, Dabezies M, Mercader VP, Krevsky B, Gatenby RA. - Nonexpandable silicone esophageal stents for treatment of malignant tracheoesophageal fistulas: complications and radiographic appearances. *Abdom Imaging.* 1997 Jan-Feb;22(1):14-9.
 4. Kishi K, Takeuchi T, Sonomura T, Kimura M, Kita K, Sato M, Terada M. - Treatment of a malignant esophageal fistula with a Gore-Tex-covered flexible nitinol stent. *Cardiovasc Intervent Radiol.* 1997 Jan-Feb;20(1):63-6.
 5. Miyata T, Watanabe M, Nagai Y, Iwatsuki M, Iwagami S, Baba Y, Furushou C, Ikuta Y, Yamamoto T, Baba H. - Successful esophageal bypass surgery in a patient with a large tracheoesophageal fistula following endotracheal stenting and chemoradiotherapy for advanced esophageal cancer: case report. *Esophagus.* 2013 Mar;10(1):27-29. Epub 2012 Jul 19.
 6. Dobrucali A, Caglar E. - Palliation of malignant esophageal obstruction and fistulas with self expandable metallic stents. *World J Gastroenterol.* 2010 Dec 7;16(45):5739-45.
 7. Stemerma DH, Caroline DF, Dabezies M, Mercader VP, Krevsky B, Gatenby RA. Nonexpandable silicone esophageal stents for treatment of malignant tracheoesophageal fistulas: complications and radiographic appearances. *Abdom Imaging.* 1997 Jan-Feb;22(1):14-9.