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## Case report

# Treatment difficulties of malignant esophagorespiratory fistula: Case report of a 56-year-old patient with esophageal cancer



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## ABSTRACT

**Introduction:** An esophagorespiratory fistula (ERF) is a lethal complication of advanced esophageal cancer. The preferred treatment method is placing a self-expanding stent, which is expected to decrease the risk of life-threatening complications.

**Aim:** In the case study we present a patient with esophageal cancer complicated with the presence of ERF, pneumonia, lung abscess and severe malnutrition.

**Case study:** A 56-year old man was hospitalized due to short syncope, dyspnea and cough. Cachexia was apparent. Immediate diagnostics with chest X-ray, bronchoscopy, gastroscopy and computed tomography (CT) of the chest revealed esophageal cancer and presence of ERF with respiratory complications. Endoscopic stent placement significantly decreased the initial symptoms. The patient was later re-admitted due to recurrent respiratory infections, dysphagia and progressing cachexia. He required stent placement again, parenteral alimentation and prolonged antibiotic therapy. From the diagnosis he survived 28 weeks.

**Results and discussion:** The average survival of patients with diagnosed ERF is about 8 weeks. The palliative treatment is expected to reduce bronchial aspirations and to prevent dysphagia. The recommended method is the insertion of esophageal stent to unblock the gastrointestinal tract and to close the fistula simultaneously. Reopening of the ERF is a severe complication caused mostly by progressing neoplasm. Successful surgical treatment of primary or recurrent fistulas is only probable in patients with good or moderate performance status.

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**Conclusions:** An immediate implementation of diagnostic and therapeutic methods is necessary, as the time to diagnosis and treatment of a malignant fistula strongly influences the patient's survival and quality of life.

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## 1. Introduction

About 450 000 new cases of esophageal cancer are registered in the world each year. The most frequent squamous-cell carcinoma (SCC) and adenocarcinoma stand for about 90% of malignant neoplasms in this location and in about 80% of cases, males aged above 40 are involved.<sup>1</sup> The risk factors are: smoking, alcohol abuse, lower social status, radiotherapy of the mediastinal area or corrosive burns of the esophagus in the past, persistent gastroesophageal reflux or head and neck tumor in the medical history.<sup>2</sup> In most patients esophageal cancer at the time of diagnosis is already in an advanced stage and excludes radical treatment. The patients undergo palliative treatment with the aim of life comfort improvement and proper nourishing if esophageal stenosis occurs. The average survival from the diagnosis time-point is several months only.<sup>3</sup>

A severe complication of esophageal cancer is an esophago-respiratory fistula (ERF) that is observed in about 1%–22% of patients with active malignant esophageal disease and in less than 1% of patients with respiratory tract neoplasms. Due to the high probability of substance aspiration through an abnormal canal from the gastrointestinal into the respiratory tract, the patients often suffer from severe pneumonia, lung abscess or sepsis. The most frequent symptoms of the fistula are cough with fever and food intolerance that can be mistaken for dysphagia in course of esophageal stenosis. The autopsy data suggest higher rate of malignant fistulas than it is actually noted in living patients.<sup>4,5</sup> The high risk of lethal complications requires an immediate but prudent intervention. The former first choice method using plastic non-expandable esophageal stents was associated with a high rate of complications (in 15%–40%): perforations, hemorrhages, compressive necrosis, obstructions and movement of the prosthesis beyond the area of its implantation. Recently it has been replaced by an endoscopic placement of a self-expanding stent that has higher effectiveness and lower rate of complications.<sup>6</sup>

## 2. Aim

In the case report we describe diagnostic and therapeutic difficulties regarding a 56-year-old patient with an advanced esophageal cancer where the ERF was already present at the time of the malignancy diagnosis.

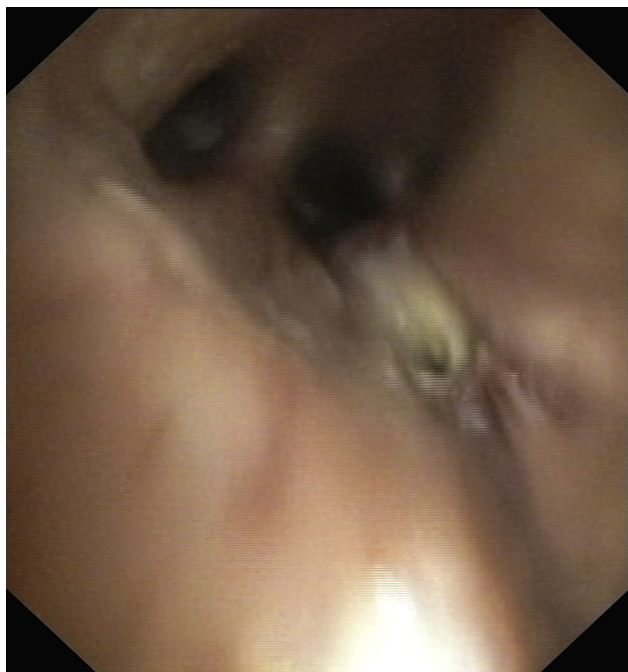
## 3. Case study

A 56-year old man with a history of alcohol and cigarettes abuse was admitted to the internal medicine department due

to a stomach pain, a short syncope without loss of consciousness and dyspnea. For several months he complained about weakness and loss of appetite, and for the preceding two weeks he suffered from vomiting and annoying cough about 30 min after each meal. Loss of 10 kg of weight was observed. In physical examination cachexia was apparent. The chest X-ray evoked a strong suspicion of a tumor in the right lung hilus (Fig. 1). An infiltration in the orifice to the middle lobe and a bronchoesophageal fistula in this area were found in bronchoscopy (Fig. 2). In gastroscopy an ulcerative infiltration starting at the 25th centimeter of esophagus was noted and additionally a clefty lumen at the 30th centimeter that periodically excreted some purulent contents (Fig. 3). In the specimen from the bronchial tree there were only inflammatory and necrotic changes observed, while in the esophageal ulceration cells of carcinoma planoepitheliale G2 were present. The chest CT scan confirmed an esophageal neoplasm in the form of a circular infiltration, spreading from the tracheal bifurcation level up to the level of lower pulmonary veins, with progression toward the mediastinum; additionally, right-sided pneumonia and a developing abscess (3 × 2 cm) in the 6th right lung segment could be observed; there was no direct fistula lumen between the bronchial and the gastrointestinal system

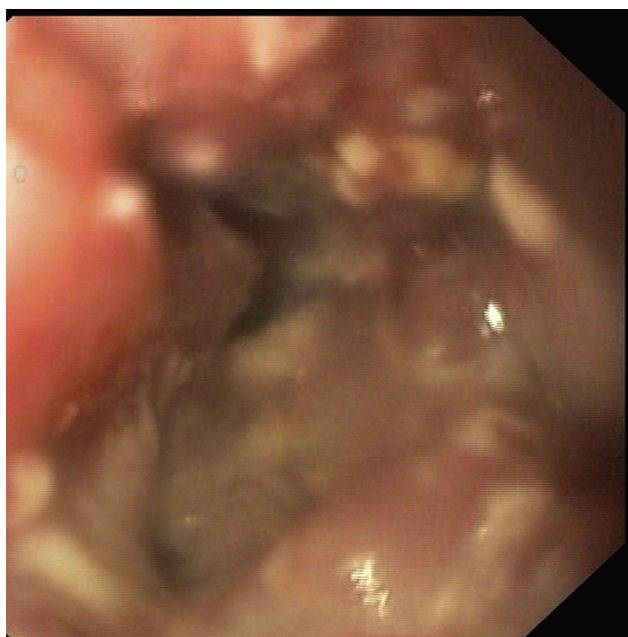


**Fig. 1 – Chest X-ray. Suspicion of a tumor in the right lung hilus.**

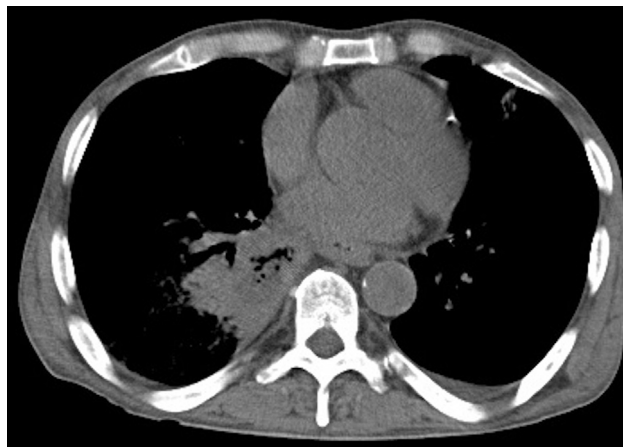


**Fig. 2 – Bronchoscopy. Bronchoesophageal fistula.**

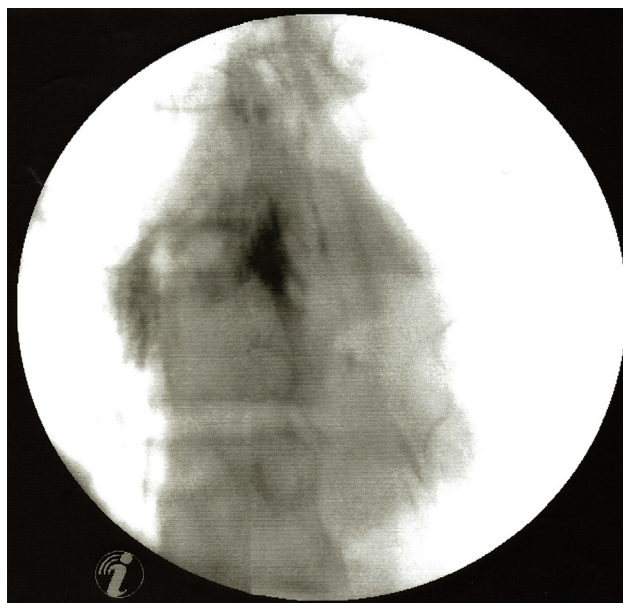
visible in CT (Fig. 4), while it was confirmed by means of fluoroscopy using contrast (Fig. 5). The patient had a partially-covered esophageal stent (Endo-Flex diameter 20 mm and length 120 mm) inserted. The procedure significantly decreased the gastrointestinal and respiratory tract symptoms. After 19 weeks from the hospital discharge the patient was re-admitted due to an upper respiratory tract infection with mild dysphagia. The chest X-ray revealed no inflammatory changes and the patient was released with oral antibiotics and directed to the



**Fig. 3 – Gastroscopy. Ulcerative infiltration of the esophagus.**



**Fig. 4 – Chest CT. Right-sided pneumonia with developing abscess.**



**Fig. 5 – Fluoroscopy. Right-sided esophagorespiratory fistula confirmed.**

outpatient clinic for alimentation. However, after a week he required hospital admission again, due to radiologically confirmed right-sided pneumonia. He had not taken the prescribed antibiotics before due to the worsening of dysphagia. Emergency gastroscopy revealed an occlusive narrowing of the esophagus at the 28th centimeter; in chest CT scan the neoplasm infiltration was closing the lumen over the upper end of the stent placed 20 weeks before (Fig. 6). Another stent was placed (Ultraflex Esophageal NG Covered 18 × 120 mm), and after the procedure the patient was able to swallow half-liquid contents. The last 4-week-long hospitalization that ended with death, took place after the next 9 days from hospital discharge. The patient was admitted due to a severe upper respiratory tract infection with dehydration, hypotension and hemoptysis. Despite intensive treatment and implementing parenteral alimentation, the clinical state of the patient was progressively deteriorating





**Fig. 6 – Chest CT. Infiltration closing lumen of the previously placed stent.**

and cachexia was increasing quickly. From the moment of diagnosis the patient survived 28 weeks.

#### 4. Results and discussion

The accepted treatment methods of malignant ERF are palliative procedures that are expected to reduce the risk of aspiration into the respiratory tract and to prevent the development of cachexia due to the restricted patency of the infiltrated esophagus. Statistical data suggest that the first choice method in patients with the coexistence of an esophagus obstruction and an ERF but without a bronchial obstruction is the insertion of a self-expanding covered esophageal stent that is supposed to unblock the gastrointestinal tract and to close the lumen of the fistula simultaneously.<sup>4</sup> Gastrostomy or jejunostomy are designed for patients in whom the esophageal stenting is not performable or previous esophageal stenting was not effective. If technically possible, operative procedures should be considered.<sup>7</sup> According to Choi et al., the average survival of patients with ERF is about 8 weeks from the time of its diagnosis.<sup>8</sup> Our patient survived almost 28 weeks, although at the diagnosis time the ERF had already been complicated with right-sided pneumonia, a developing abscess and progressing cachexia. In literature, survival over a year can be found, but it refers to patients who were surgically treated.<sup>9</sup> The development of a fistula is mostly caused by neoplasm progression, rarely it is a complication of cancer treatment such as surgical resection, esophageal and bronchial stenting, esophagus skipping anastomosis, chemotherapy, radiotherapy or laser therapy. Only in a few cases the first symptoms of esophageal cancer depend on the presence of a fistula or its complications as it was in our patient. He was disqualified from chemotherapy and radiotherapy that are in general contraindicated for patients with the ERF because the oncological treatment can cause its progression or enlargement due to the tumor cell necrosis. Implementing only a single method of palliative treatment is often ineffective, because the rapidly growing neoplastic mass can obstruct the residual lumen of the esophagus and require further surgical procedures.<sup>10</sup> Considering the case patient, re-implantation of an esophageal stent was

required just 22 weeks after the first stent placement, because of the upper occlusion of gastrointestinal tract that was radiologically confirmed. On the other hand, the esophageal re-stenting to improve the enteral alimentation seems to have a lower rate of complications than f.e. percutaneous endoscopic gastrostomy (PEG).<sup>11</sup> According to Adler et al., the placement of PEG tubes through previously placed esophageal stents is connected with a higher rate of gastrostomy tube impaction within the stent and resultant stent migration.<sup>12</sup> Regarding our patient, it is of importance that in the course of re-hospitalizations there was no endoscopic or radiological evidence of recurrence of the fistula, despite recurring respiratory tract infections. According to Shin et al.,<sup>4</sup> a re-opening of the ERF is a life threatening condition that was observed in 35% of patients who had undergone esophageal stenting and was caused mostly by the rapidly growing and infiltrating neoplastic mass. Successful treatment of recurrent fistulas is only probable in patients in a good or moderate clinical state, which is a rare phenomenon considering the fact that the fistula is a symptom of a highly aggressive malignancy.<sup>13</sup> In case of severe dyspnea or other symptoms of respiratory tract stenosis, parallel bronchial stenting should be considered.<sup>14</sup> In the described case, parenteral alimentation was implemented due to the increasing cachexia, although according to Balazs et al., such a treatment does not influence the survival time but improves the quality of life only.<sup>15</sup>

#### 5. Conclusions

An ERF should be suspected in every patient with esophageal cancer who presents recurrent cough after meals, or in whom severe pneumonia or a lung abscess is diagnosed. The reported case proves the necessity of immediate implementation of various diagnostic methods and treatment. Although the proposed treatment can only improve the patient's life comfort without influencing the survival, in the reported case the overall survival was significantly longer than it could be prognosticated at the time of the diagnosis of esophageal cancer with an esophagorespiratory fistula.

#### Conflict of interest

The authors declare no conflict of interest.

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