

## Late Epiphrenic-Retroperitoneal Fistula of an Esophageal Diverticulum Ten Years after Surgical Excision: Report of the First Case

Fabbri Nicolò\*, Ferro S, Pansini G, Quarantotto F, Anania G, Carcoforo P and Cavallesco G

Department of Morphology, Experimental Medicine and Surgery, Section of General Surgery, University of Ferrara, Ferrara, Italy

\*Corresponding Author: Fabbri Nicolò, Department of Morphology, Experimental Medicine and Surgery, Section of General Surgery, University of Ferrara, Ferrara, Italy.

Received: August 29, 2019; Published: September 16, 2019

### Abstract

**Background:** Esophageal fistula is a rare complication of an epiphrenic diverticulum and represents a difficult diagnostic challenge. In the majority of cases in the English literature, the fistula is secondary to the spontaneous perforation of the epiphrenic diverticulum and in only one case an esophagobronchial fistula occurs after resection of an epiphrenic diverticulum. No case of esophageal fistula communicating with the retroperitoneum tissues was found.

**Case Report:** We present a case of 84 years old male admitted in 2017 in our Hospital for the appearance of epigastralgia and cough with traces of blood to sputum, asthenia and night sweats. Ten years before he underwent to a hiatal hernia plastic sec. Nissen and subsequently was reoperated for exeresis of diverticulum of the distal third of the esophagus by thoracotomy. In August 2008, because of esophageal substenosis an operation of posterior gastropexy sec. Hill with the demolition of the previous hiatoplasty was performed. In 2017, a computerized tomography shows a voluminous abscess in the right paravertebral region with development along the muscular plane of the ipsilateral psoas and a fistular path of about 4 cm between the posterior wall of the esophagus and the retroperitoneal collection of the abdomen.

**Conclusions:** Esophageal fistula represents a complex problem of epiphrenic diverticulum and rarely a hidden complication of surgery. Surgery is the treatment of choice in suitable patients.

**Keywords:** Epiphrenic Diverticulum; Esophageal Fistula; Abdominal Wall

### Introduction

Esophageal diverticula are evaginations of the mucous projecting from the lumen of the esophagus. Traction diverticulum is an external inflammatory reaction in neighboring mediastinal ganglia that adhere to the esophagus and retract the wall in a way that contracts and scars the tissue, provoking a true diverticulum, meaning that it has as many layers as the esophageal wall itself. This rarely shows any symptoms, and in this case, no treatment was deemed necessary. Traction esophageal diverticula result from an inflammatory contracture outside the esophagus. An epiphrenic oesophageal diverticulum develops as the result of increased pressure in the oesophageal wall (pulsion diverticulum). Epiphrenic diverticula occur in the lowermost 10 cm of the esophagus. The main symptoms are dysphagia, regurgitation, and pain when swallowing food [1]. Radiological studies indicate that the prevalence of epiphrenic diverticula in the general population is 0.015 % in the USA [2] and in a more selective dataset from Europe, epiphrenic diverticula were found in 2 % of patients who were examined for swallowing disorders [3].

The largest single-case series to date only included 57 patients over a 15-year period [4].

The original surgical procedure was left side thoracotomy with dissecting free and resection of the diverticulum [5]. In cases of concomitant achalasia or increased pressure in the lower oesophageal sphincter, it is increasingly common to perform myotomy of the esophagus on the opposite side of the diverticulum and an antireflux operation, normally with partial fundoplication, to counteract acidic and alkaline gastro-oesophageal reflux. Myotomy on the opposite side of an extirpated diverticulum reduces the risk of leakage compared with excision without myotomy [6,7]. The in-hospital mortality of this type of surgery rate is 5.9% [7]. Esophageal fistula is a rare complication of an epiphrenic diverticulum and represent a difficult diagnostic challenge. In a recent literature review, a total of 35 cases had been reported. In 28 misdiagnosed cases, lung infection was the most frequently diagnosed disease (19 cases), followed by bronchiectasis (6 cases) and pulmonary abscess (4 cases) [8].

We performed research using Pubmed and searched all articles in English literature containing “esophageal diverticulum” and “fistula” in the title. 14 English articles were found and in only one case an esophagobronchial fistula after resection of an epiphrenic diverticulum was found. In an article of 2014 Hauge et al reports the casistic of Oslo University where only one case who had undergone reoperation for leakage and oesophageal mediastinal fistula.

In the other cases found the esophageal fistula was secondary to the spontaneous perforation of the epiphrenic diverticulum. No case of esophageal fistula communicating with the retroperitoneum tissues was found.

## Case Report

In 2017 an 84 years old male was admitted in Hospital for the appearance of epigastralgia and cough with traces of blood to sputum, asthenia and night sweats. In the anamnesis, there was a history of ischemic heart disease, atrial fibrillation in anticoagulant treatment, colic diverticulosis and erosive gastritis, hypertension, previous cholecystectomy. chronic renal failure with nephroangiosclerosis, diverticulosis of the colon, COPD. The patient had undergone the operation of hiatal hernia plastic sec. Nissen and subsequently reoperated for exeresis of diverticulum of the distal third of the esophagus with the size of 10 cm with a large collar by thoracotomy. The esophagus radiograph shows marked stenosis of the esophageal-gastric junction with the suspicion of a small fistula 2 cm long above the esophago-gastric junction with the anterior course which is not further identified after a week with the repetition of the examination.

In August 2008, because of esophageal substenosis an operation of posterior gastropexy sec. Hill with the demolition of the previous hiatoplasty was performed.

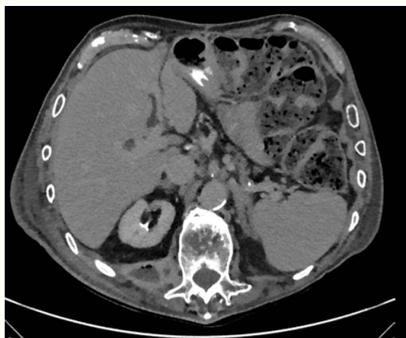
One year later (September 2009) the patient presented to the emergency room for epigastric pain, profuse sweating and diarrheal discharge with melena. the electrocardiogram excludes acute ischemia, the blood counts indicate an anemization (Hb 9.4 g dl) which is treated with martial therapy.

The patient was investigated with computed tomography in the following years after surgery due to the presence of bilateral basal consolidation areas of the lung parenchyma of non-univocal interpretation. In March 2014, the patient is again rescued for dyspnea, epigastralgia, and white blood cell elevation. On this occasion, bronchopneumonia is diagnosed and treated effectively with antibiotic therapy.

In 2017 the patient is admitted to the hospital for a cough with hemoptysis, severe asthenia, and night sweats, production of purulent catarrh and sensation of weight at the gastric level. Therefore, he performs esophagogastrosocopy that describes the presence of probable esophageal pseudo diverticulum apparently closed by suture, 38 cm distant from the oral line and esophagitis. A computerized tomography of the chest with diagnostic completion describing a reduction of bilateral basal pulmonary consolidation area was performed.

At the end of November 2017, the patient repeated a computerized tomography of the thorax with evidence of a dimensional increase of the abscess in the right paravertebral region with extension along the muscular plane of the ipsilateral psoas with a diameter of about 8 centimeters.

The presence of pseudo-esophageal diverticulum with fistula was therefore suspected.



**Figure 1:** Computerized tomography with harvesting presence with air bubbles in the right diaphragmatic subset.

Due to the worsening of clinical conditions, the patient was subjected to incision of abscess collection in the right flank with abundant purulent material and a thoracentesis due to the presence of abundant reactive pleural fluid was performed. At the CT scan, it was possible to appreciate the presence of a fistular path of about 4 cm between the posterior wall of the esophagus and the retroperitoneal collection of the abdomen.



**Figure 2:** Presence of a fistular path of about 4 cm between the posterior wall of the esophagus and the retroperitoneal collection of the abdomen.

The patient kept fasting from food and liquids using parenteral nutrition. Subsequently, in January 2018 a jejunostomy was placed to start enteral nutrition while maintaining the esophagus-gastric tract at repose.

The wound on the right side of the abdomen was medicated daily by placing a medicated sapphire with marked improvement of local soft-tissue flogosis. An EGDS is performed with identification of esophageal orifice on which a Pad look clip is placed. However, the presence of peri-fistulous fibrinous tissue does not allow it to be kept in place. A second unsuccessful attempt is made by positioning Resolution clip. The patient was subjected to numerous thoracentesis by aspirating citrine reactive liquid and culture test with identification of Escherichia coli ESBL positive which was treated with Tazocin and Clindamycin and Candida Albicans for which an antifungal therapy is

set with fluconazole 400 milligrams twice a day for two days then fluconazole once a day for 3 days and subsequently modified with Candidas 50 milligrams 2 vials on the first day and subsequently 1 vial die and Zerbaxa 1 gram intravenous 3 times a day with improvement of laboratory tests and reduction of the C reactive protein.

The CT scan performed after 2 weeks describes a clear reduction in the collection in the thoracic and paravertebral area. About 1 month after positioning the jejunostomy in the left side of the abdomen it was necessary to remove it due to the appearance of an infected fluid collection in the subcutaneous area that required its surgical drainage. The patient gradually resumed his recovery and was discharged at the end of February. He performed weekly wound dressings and Vacuum Assisted Closure Therapy (Vac Therapy) was set. The last contact dates back to July 2018 due to fistula medication. The patient subsequently died of other causes.

**Discussion**

The esophageal fistula represents a rare complication in the diverticulectomy of the esophagus. There are few scientific papers in the literature that report cases of esophagus-bronchial or esophagus-pleural fistula, while there are no cases of fistulas between the esophagus and the retroperitoneal region as in our case. The following table 1 shows the cases still present in the literature, available on the Pubmed database.

Authors	Side of fistula	Sex	Age	Time after surgery	Therapy
Bonavina, <i>et al.</i> [9]	Recurrent epiphrenic diverticulum and esophago-bronchial fistula	male	54	3 years	Surgical correction (transhiatal stapling of the pouch combined with distal esophageal myotomy and Dor fundoplication).
Tobias Hauge, <i>et al.</i> [1]	fistula from the oesophagus to the mediastinum	ND	ND	ND	Fistulectomy 1 year later
Fabbri, <i>et al.</i>	Esophagus-retroperitoneal fistula after epiphrenic diverticulum resection and iato plastic surgery	male	84	10 years	Conservative treatment

**Table 1:** Cases of esophagus fistula after epiphrenic diverticulum resection.

Probably the symptoms reported by our patient in the first years after surgery represented the first signs of complication, however it was not possible to highlight the existence of the fistula. The long latency between the surgery and the appearance of fistulized retroperitoneal abscess in the skin plane has probably influenced the scarce efficacy of the conservative treatments implemented, as it allowed the stabilization of the fistulous path. In these situations the patient should be operated to remove the fistula, however, in our case, it was not possible due to advanced age, the numerous comorbidities and the extreme anatomical complexity of this case. The positioning of clips by endoscopy was ineffective due to the presence of fibrinous tissue in the vicinity of the esophageal fistulous orifice which did not allow adequate stability. Furthermore, the site of the fistula near the cardia, the presence of a residual pseudo diverticulum with angulation of the distal esophagus and the previous surgical operations performed did not make it possible to place a stable esophageal stent.

Finally, the VAC therapy allowed to keep the wound more cleansed and it was possible to reduce the frequency of outpatient medications after discharge. The patient subsequently died by other causes.

**Conclusion**

The esophageal fistula represents a rare but dangerous complication of the esophageal diverticulum with an insidious and often sub-clinical onset. These features can delay diagnosis for several years from the first symptoms. it can rarely occur after diverticulectomy.

Treatment should be surgical when it is possible, by removing the fistula. Conservative treatment should include prolonged fasting, endoscopic clip placement, and adequate antibiotic therapy. Unfortunately, the chronic nature of the fistula makes it difficult to resolve.

### Acknowledgments

None.

### Bibliography

1. Hauge T, *et al.* "Surgical treatment of epiphrenic oesophageal diverticulum". *Tidsskrift for Den norske Legeforening* 134.10 (2014): 1047-1050.
2. Wheeler D. "Diverticula of the foregut". *Radiology* 49.4 (1947): 476.
3. Schima W, *et al.* "Association of midoesophageal diverticula with oesophageal motor disorders". *Acta Radiologica* 38.1 (1997): 108-114.
4. Macke RA, *et al.* "Thoracic esophageal diverticula: A 15-year experience of minimally invasive surgical management". *Annals of Thoracic Surgery* 100.5 (2015): 1795-1802.
5. Soares R, *et al.* "Epiphrenic Diverticulum of the Esophagus. From Pathophysiology to Treatment". *Journal of Gastrointestinal Surgery* 14.12 (2010): 2009-2015.
6. Zaninotto G, *et al.* "Therapeutic strategies for epiphrenic diverticula: Systematic review". *World Journal of Surgery* 35.7 (2011): 1447-1453.
7. Chan DSY, *et al.* "Systematic Review and Meta-analysis of Surgical Treatment of Non-Zenker's Oesophageal Diverticula". *Journal of Gastrointestinal Surgery* 21.6 (2017): 1067-1075.
8. Zuo CY and X Z. "Esophageal bronchial fistula secondary to esophageal diverticulum: A case report and literature review". *Zhonghua Jie He He Hu Xi Za Zhi* 41.8 (2018): 622-627.
9. Bonavina L, *et al.* "Esophagobronchial fistula after thoracoscopic resection of an epiphrenic diverticulum". *Diseases of the Esophagus* 12.4 (1999): 324-325.

Volume 6 Issue 10 October 2019

©All rights reserved by Fabbri Nicolò, *et al.*