



## Stent-In-Stent Treatment of Malignant Esophagopericardial Fistula: A Successful Multidisciplinary Approach

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### Abstract

The Authors present a tangled case of malignant esophagopericardial fistula complicated by pneumopericardium, sepsis, and hemodynamic instability in which a multidisciplinary assessment has achieved a positive clinical result by a permanent stent-in-stent esophageal technique.

**Keywords:** Fistula; Pericardium; Esophagus; Tomography

### Introduction

Esophagopericardial fistula is an unusual and potentially life-threatening complication caused by several pathologies of the esophagus [1-3]. Often the fistula is iatrogenic, following esophagoscopy or esophageal irradiation or, less frequently, traumatic due to ingested foreign body, as fish bone or caustics. Recently, the widespread use of radiofrequency ablation procedures for treatment of atrial arrhythmias has led to a significant increase in this potentially devastating complication [4]. More rarely fistulas complicate an esophageal neoplastic disease [5]. The overall mortality rate is very high, reaching 80% in malignancies [6].

### Case Presentation

A 68-year-old woman with 1-year medical history of esophageal squamous cell carcinoma with liver metastases (neoplasm staging IVb: T4, N1, M1 as defined by American Cancer Society, 2020) was admitted to the emergency room due to hemodynamic instability, orthopnea and chest pain. Nine months before, an implantation of partially covered Self-Expanding Metal Stent (SEMS, Evolution PC Cook Medical Inc, Bloomington, IN) has been performed, as a palliative treatment of a malignant esophageal stricture. At admission, chest X-ray demonstrated a conspicuous hydropneumopericardium (Figure 1), and the patient was referred to our cardiology department. A physical examination revealed arterial hypotension (88/55 mmHg), sinus rhythm (96 bpm), and tachypnea (respiratory frequency 32/m). Blood tests showed leukocytosis (23.150/mm<sup>3</sup>) with neutrophilia (91%) and elevated C-reactive protein (32.12 mg/dL). No fever was present. Urgent 2-D transthoracic echocardiogram showed a very large circumferential hyperechoic pericardial effusion with moderate compression of the right atrium. Pericardial drainage was immediately

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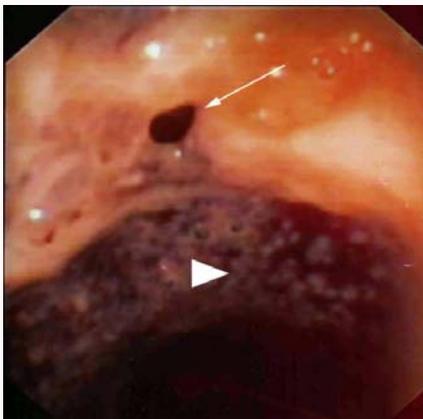
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**Figure 1:** Chest X-ray at admission (anteroposterior and lateral projections). Grossly dilated pericardial sac with air-fluid level (hydropneumopericardium) and evident pericardial thickening. The lateral X-ray projection shows the previously implanted esophageal stent (arrows).



**Figure 2:** Chest computed tomography scan with Gastrografin® transit shows massive pneumopericardium (asterisks) and irregularly thickened pericardial leaflets. Free leakage of Gastrografin® within the pericardial space (arrow) reveals the presence of a fistulous tract. Gastrografin® fills the esophageal stent (arrowhead).



**Figure 3:** Esophagoscopy shows the perforation of the mid-thoracic esophagus (arrow) and the irregularity of the esophageal mucosa. Cardiac pulse was visible through the ulcerated perforation. The arrowhead indicates the partially covered proximal flanges of the previous implanted stent.

performed by subxiphoid approach under fluoroscopic guidance with a pigtail catheter by means of the Seldinger technique. A volume of approximately 650 ml of cloudy and foul-smelling fluid was drained. The catheter was left in place for 4 days for continuous drainage and repeated flushing with saline. Pericardial fluid analysis was performed and results were suggestive of salivary flora colonization consisting of numerous Gram-positive and Gram-negative bacilli. Broad-spectrum antibiotics were started. The clinical suspicion of an underlying esophagopericardial fistula was confirmed by thoracic Computed Tomography (CT) with oral iodinated media contrast (Gastrografin® Bayer). CT scan visualized the fistulous tract and the leakage of contrast from esophagus into the pericardial cavity, excluding other esophageal fistulation (Figure 2). The subsequent flexible fiber-optic esophagoscopy showed, 34 cm distal to incisor teeth's, a crater-like ulceration on the anterior wall of the mid-thoracic esophagus, with an evident perforation penetrating into the pericardial space, confirming the presence of an esophagopericardial fistula (Figure 3). The sharing of the clinical case with the oncologist and the gastroenterologist defined the most appropriate diagnostic path. We scheduled our patient for palliative therapy, due to advanced neoplastic disease and the rapid worsening of clinical condition. A fully covered SEMS (Evolution FC Cook Medical Inc, Bloomington, IN) was placed by "stent-in-stent technique", with successful sealing of the fistula. Control esophagogram with Gastrografin® revealed normal flow of



**Figure 4:** Control esophagogram with Gastrografin® revealed normal flow of contrast through the esophagus into the stomach without evidence of the esophageal fistulation.



**Figure 5:** Chest RX controls (2 months after stent-in-stent procedure) shows normal cardiac shape and no recurrent pericardial effusion.

contrast through the esophagus into the stomach without evidence of the fistula (Figure 4). Both implanted stents were left in place and no further interventional procedures were performed. The patient has been taking oral soft nutrition 4 days after the procedure. Chest RX control two months after stent placement shows normal cardiac shape and no recurrent pericardial effusion (Figure 5). During clinic and endoscopic follow-up no further complications of the esophageal canalization, stent dislocation or embolic and septic events occurred, until she died six month later, due to her neoplastic disease.

## Discussion

The case highlights a critical and complicated context that required a multidisciplinary approach to successfully face and resolve this intricate clinical situation. The multidisciplinary team, including the intensive care cardiologist, the senior digestive endoscopist, the oncologist and the radiologist, discussed the clinical case, considered the patient's choices, and planned a tailored care plan. The first step was the resolution of the severe septic state and the drainage of the pericardial space, cause of the hemodynamic imbalance. The second step was the choice of the definitive treatment of the esophagopericardial fistula. The management of esophagopericardial fistula depends on the underlying pathogenesis [7,8]. Surgical approach is mandatory for

nonmalignant fistulas, although alternative nonsurgical approaches are reported in the literature, as esophageal stent placement [9]. When surgery is not indicated or refused, the palliative treatment by esophageal stent placement provides a valid alternative, ensuring improvement of patient's quality of life as well as fast resumption of oral nutrition, and minimally morbidity [10,11]. Our concept was to combine a minimally invasive nonsurgical approach by esophageal stent placement with resolution of septic status and pericardial effusion by continuous drainage, repeated flushing with saline and systemic broad-spectrum antibiotic therapy. In our patient a partially covered SEMS had previously been implanted for an esophageal neoplastic pathology. The senior endoscopist indicated the palliative implantation of a fully covered SEMS inside the first partially covered SEMS by stent-in-stent technique [12,13]. This technique is usually performed for the removal of stents embedded in the esophageal wall. Normally, one or both stents were subsequently removed after a few weeks. However, the removal of stents is frequently associated with bleeding or mucosal tears due to removal of the superficial layer of the esophagus or with mechanical complications due to traction of device [12]. For these reasons, and for the severe underlying pathology, it was decided not to proceed with removal maneuvers [13]. This case highlights the importance of the multidisciplinary approach that conveys benefits to both the patients and the health professionals working on the team, improves clinical outcomes and offers enhanced safety for patients, by a decision making from different specialty fields that lead a shared treatment strategy patient-centered.

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## Author Contributions

Guarantors of integrity of entire study, D.A.; manuscript drafting or manuscript revision for important intellectual content, D.A., C.C.; approval of final version of submitted manuscript, all authors; literature research, D.A.; and manuscript editing, D.A., C.C.

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