Aortic-Esophageal Fistula (AEF) - A Case Report

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Introduction

An aortoesophageal fistula is a life-threatening cause of gastrointestinal bleeding where an abnormal communication between the esophagus and the aorta may result from a thoracic aortic aneurysm, foreign body ingestion, esophageal malignancy, or postoperative complications. The diagnosis can be made on the basis of clinical findings alone. (Heckstall & Hollander, 1998). Diagnosis of AEF is rarely made before massive hematemesis. However, most cases are associated with characteristic Chiari’s triad features of chest pain, hematemesis, symptom-free period after hematemesis. However, the triad is expressed only in 1/3 of patients. The leading diagnostic tools are esophagogastrroduodenoscopy (detection accuracy ≈ 25%) and contrast CT angiography, which in most cases reveals the disease. After the diagnosis, the patient’s aorta and esophagus are urgently reconstructed by the endovascular method. Massive bleeding from the esophagus, diagnostic difficulties, and the insufficient amount of medical equipment in the regions of Georgia make it difficult, and sometimes the possibility of diagnosing the above-mentioned disease is missed altogether, and thus complicates the solution. We present a case of aorto-esophageal fistula, the patient was brought to our clinic from one of the regions of Georgia, where he was being treated with the following diagnosis: bleeding from an esophageal ulcer. In our hospital the patient was immediately diagnosed by computed tomography (CT).

Abstract

Aortoesophageal fistula is one of the rarest causes of bleeding in the upper gastrointestinal tract. Aneurysm of the thoracic aorta is the leading causing factor in such cases. The main challenge for doctors is to diagnose timely, because the disease can be accompanied by massive bleeding and rapid worsening of the patient’s condition. A characteristic symptom is Chiari’s triad (chest pain, hematemesis, symptom-free period after hematemesis). However, the triad is expressed only in 1/3 of patients. The leading diagnostic tools are esophagogastrroduodenoscopy (detection accuracy ≈ 25%) and contrast CT angiography, which in most cases reveals the disease. After the diagnosis, the patient’s aorta and esophagus are urgently reconstructed by the endovascular method. Massive bleeding from the esophagus, diagnostic difficulties, and the insufficient amount of medical equipment in the regions of Georgia make it difficult, and sometimes the possibility of diagnosing the above-mentioned disease is missed altogether, and thus complicates the solution. We present a case of aorto-esophageal fistula, the patient was brought to our clinic from one of the regions of Georgia, where he was being treated with the following diagnosis: bleeding from an esophageal ulcer. In our hospital the patient was immediately diagnosed by computed tomography (CT).
aortoesophageal syndrome, including chest pain and sentinel hematemesis of red blood followed at a variable interval of time by rapidly fatal massive exsanguinating hematemesis. In the present series these symptoms were observed in 4 of the 6 patients with true AEF. (Kieffer, Laurent, & Dominique, 2003). CT angiography is the mainstay for diagnosis owing to the quick acquisition, widespread availability, and superior spatiotemporal resolution (Vu, Menias, & Bhalla, Radiographics). Extravasation of contrast material from the aorta into the esophagus, or vice versa, can be diagnosed with high confidence by CT angiography.

**Pathogenesis**

Atherosclerotic disease leading to intimomedial fat deposition is the most common cause of loss of integrity of an otherwise intact vessel wall. With the high pulsatile intravascular pressures, this phenomenon gradually leads to aneurysmal growth. The weakened aortic wall allows abnormal radial transmission of pulsatile pressure to the surrounding soft tissues. This abnormal force causes pressure necrosis and adhesive granulation tissue between the aorta and a periaortic hollow or solid organ, resulting in a fistula (Picichè, R, & A, 2003). A false aneurysm, or pseudoaneurysm, does not contain wall layers and is more prone to sudden rupture instead of controlled fistula development. A true aneurysm occurs due to weakening of the wall of an otherwise intact blood vessel, in which case all three walls of the artery are thinned but intact. If AEF (Aortic-esophageal Fistula) occurs during aortic intervention and/or aortic graft placement, it is termed a secondary fistula. The pathophysiology of secondary fistulas is multifactorial and related to foreign body reaction to the graft, graft cramping, infection, leakage coil placement, etc.

**Case Presentation** – A 55-year-old male patient was transferred from one of the regional clinics of Georgia, where he was being treated with the diagnosis - esophageal ulcer, thoracic aorta aneurysm without rupture. Bleeding relapses were noted twice before transferring, with vomiting of bright red blood for which a Blakemore tube was inserted. Hemotransfusion took place. Due to the pain in the chest, a computed tomography of the chest cavity with I.V contrast was performed, where a 1.6/2.3 cm contrast-filled mass was noted on the right wall of the aorta, after which the penetration of the aorta into the esophagus was suspected.

Upon admission, he complains of general weakness, dizziness, shortness of breath, chest pain, the patient is drowsy, opens his eyes when called, and is able to perform simple tasks. 5 years ago, left inguinal hernia plastic surgery was performed, there is no allergy. Due to the general condition, the patient was intubated. The condition is stable, a preliminary diagnosis was made:

- Thoracic aortic aneurysm, ruptured – I71.1
- Somnolence, stupor and coma – R40
- Other shock – R57.8
- Acute respiratory failure – J96.0
- Essential hypertension – I10

**Preoperational Condition**

The blood shows an anemic picture, post-hemorrhagic anemia, which is why blood transfusion became necessary. In the background of high CRP, the blood is sterile. During the ultrasound examination of the upper part of the abdominal cavity, a large amount of mixed echogenic mass - blood - is visualized in the stomach with a Blakemore tube. It was decided to perform an integrated operative intervention, to insert stent grafts of the aorta and esophagus. In order to avoid the infection of the aortic stent graft, the esophageal stent graft should be inserted first, and then the...
Interventions on the aorta must be performed. Recent studies showed that stent grafting is a promising technique for treatment of esophageal perforation. However, the evidence of its benefits is still scarce. Stent grafting seems to be an effective less invasive technique for the treatment of esophageal perforation. It allows the preservation of the esophagus in most of patients. (Biancari, Tauriainen, & Tatu, 2017).

**Research Conclusion**

Fifty-three consecutive patients underwent stent grafting for esophageal perforation at Oulu University Hospital, Finland. The primary endpoint of this study was early and intermediate mortality. Secondary outcome endpoints were the need for esophagectomy and additional surgical procedures on the esophagus and extraesophageal structures. Patients' mean age was 64.6 ± 13.4 years. The mean delay to primary treatment was 23 ± 27 h. The most frequent cause of perforation was Boerhaave's syndrome (46.5%). The thoraco-abdominal segment of the esophagus was affected in 58.1% of cases. Minor primary procedures were performed in 25 patients (58.1%) and repeat surgical procedures in 23 patients (53.5%). Repeat stent graftings were performed in 22 patients (50%). Two patients (4.7%) underwent esophagectomy, one for unrelenting preprocedural stricture of the esophagus and another for persistent leakage of a perforated esophageal carcinoma. The mean length of stay in the intensive care unit was 6.0 ± 7.5 days and the in-hospital stay was 24.3 ± 19.6 days. In-hospital mortality was 4.6%. Three-year survival was 67.2% (Biancari, Tauriainen, & Tatu, 2017).

Stent Graft Implantation – Before esophageal stenting is started. Under general anesthesia we isolated the femoral artery and passed the extra-stiff (Lunderquist) wires into the thoracic aorta to be ready in case of bleeding. After this using endoscope esophagus lumen is free. Large blood clots and fresh blood in the lumen. At 28 centimeters from the mouth, there is an ulcerated area of up to 1 cm. Esophageal stenting was performed under X-ray control by inserting a 24-Fr esophageal stent of Cook. The penetrated area is closed without complications (Figure 3).

It was necessary to mark the exact location. For this, endoscopists infiltrated the distal site of the esophagus with contrast to make the perforation area more visible (Figure 4).

The endoscopist was then free to perform esophageal stent-grafting (Figures 5, 6).

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**Figure 3:** X-ray, Coronal plane. A – Extra-stiff wire (Lunderquist). B – Endoscope to clarify and observe the exact localisation of the perforation.

**Figure 4:** X-ray, Coronal plane. A – The contrast infiltrated area for the better measurement of the perforation.

**Figure 5:** X-ray, Coronal plane. A – 24Fr esophageal
Part of Vascular Surgery

Now is the time for vascular surgeons to act. The left common femoral artery was isolated and clamped on tourniquets. The common femoral and left brachial arteries were cannulated using the Seldinger technique. A hydrophilic wire 0.35” and an angiographic catheter “Pigtail” were transferred from the thigh in the direction of the aortic arch. Aortography was performed. A saccular aneurysm of the thoracic aorta can be seen (Figure 7).

From the femoral approach, an extra-stiff wire was passed in the direction of the aortic arch and brought to the ascending aorta. A stent-graft (40x170 mm) was transferred to the same wire. For control, a catheter is inserted from the left brachial artery and the “Pigtail” is brought to the exit of the left subclavian artery. Angiography shows a cone-shaped pathological dilatation of the left subclavian artery (Figure 8). The graft was placed in the distal descending direction of the left subclavian artery. The stent-graft is deployed. The rupture site was covered. Aneurysm is not contrasted by control angiography. Extravasation does not occur (Figure 9).

Conclusion

The patient was discharged from the hospital on 31/12/2022, with the following prescription: double anticoagulation, low dose...
Rivaroxaban + long-term aspirin, symptomatic treatment and angiolist supervision. Endoleak is not visible on repeated control computed tomography. The patient spent 5 days in the ICU and 11 days in the surgical hospital. In a nutshell, we can clearly say that this case is an obvious example of the benefits of endovascular surgery as a minimally invasive technique. In the future, similar procedures will become more and more available and will replace open surgical interventions. It is worth noting that similar and more useful tools are being created today, which helps to achieve better results. However, it should be noted that 2 weeks after discharge, the patient returned to the clinic by ambulance with signs of mediastinitis, which is why he was handed over to thoracic surgeons and is still being treated.

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Conflict of Interest: None
Ethical Considerations: None

References