

# Aortoesophageal Fistula Complicating the Aneurysm of the Descending Thoracic Aorta: a Case Report

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## Summary

**Introduction.** Aortoesophageal fistula is a rare condition defined as an abnormal connection between the aorta and esophagus. Due to the risk of massive upper gastrointestinal bleeding, aortoesophageal fistula is life-threatening and has low survival rate. Although Chiari's triad is considered a classic presentation, it appears in fewer than half of aortoesophageal fistula cases, and a definitive diagnosis is often made post-mortem.

## INTRODUCTION

Aortoesophageal fistula (AEF) is an abnormal, direct communication between the aorta and the esophagus [1, 2]. Although upper gastrointestinal bleeding (UGIB) is a common problem in daily clinical practice [3], less than 3.5% of these cases are caused by aortoenteric fistulas, including AEF [4]. Among aortoenteric fistulas, AEF accounts for around 10% [5].

AEF is classified into primary and secondary [2, 6]. Primary AEF is characterized by a spontaneous connection between the esophagus and aorta [2]. Secondary AEF arises following the treatment of aortic disease or after esophageal procedures [6]. The reported incidence of AEF varies from at least 0.007 per million people per year, up to 0.07% in the general population [7, 8]. Secondary AEF develops in roughly 1.5–2% of cases following thoracic endovascular aortic repair (TEVAR) [9].

AEF is a life-threatening condition that requires urgent diagnosis and therapeutic interventions [6]. The fatal outcome without treatment is up to 100% [10, 11], while with surgical treatment it remains up to 77% [11]. However, AEF is difficult to diagnose [12, 13]. Some patients are asymptomatic, making

**Case report.** A 65-year-old man who was found dead in his apartment. Multiple bloodstains were observed on the man's face and floor near the body at the scene. The autopsy revealed aortic atherosclerosis, a dissecting aneurysm of the descending thoracic aorta, and a 3 cm vertical aortoesophageal fistula located 5 cm below the entry to the left subclavian artery. Blood clots (1100 g) with fibrin deposits were found in the stomach, indicating an earlier separate bleeding event.

**Discussion and conclusions.** Aortoesophageal fistula is a life-threatening condition. This case underscores the challenges of promptly diagnosing aortoesophageal fistula and highlights the vital role of autopsy in revealing the cause of death in such rare conditions. In clinical practice, monitoring and managing thoracic aortic aneurysms is essential for reducing the risk of developing the aortoesophageal fistula.

**Keywords:** aortic aneurysm, aortoesophageal fistula, autopsy, bleeding, forensic medicine.

the condition challenging to suspect [14]. The diagnosis is usually confirmed only at autopsy after death [4, 15].

A rare case of sudden death due to a dissected thoracic aortic aneurysm complicated by AEF and bleeding into the gastrointestinal tract is presented below.

## CASE REPORT

A 65-year-old man was found on the floor of the bedroom of an apartment. The man was pronounced dead, and resuscitation was not initiated due to the presence of post-mortem changes in the body. The estimated post-mortem interval was 6–9 h. Multiple bloodstains were observed on the man's face and floor near the body. However, no external injuries, which could be the source of severe external bleeding were visible.

According to the medical records, the patient visited the hospital three years before his death due to bronchitis. A chest radiograph incidentally revealed a sclerotic aortic arch and a diagnosis of hypertensive heart disease was noted. There were no records of further follow-up or treatment.

An autopsy was performed to determine the cause of death. Livor mortis was visible on the dorsal surfaces of

the body and face. Bloodstains were present on the face, around the nostrils, and the oral cavity, as well as on the anterior surface of the chest, abdomen, and inner surface of the left hand. A small,  $1.2 \times 2$  cm, perimortem superficial abrasion of the skin was observed on the forehead above the left eyebrow. No other external injuries were observed.

Internal examination revealed significant atherosclerosis of the aorta with calcified and ulcerated atherosclerotic plaques covering more than half of the aortic inner surface area. A 16 cm long dissected aneurysm with a diameter of up to 8 cm was identified in the descending thoracic aorta (Fig. 1). A yellowish-white, firm, pear-shaped fibrin clot was observed between the outer and inner walls of the aorta. In the middle third of the aneurysm, the aortic wall was partly covered by a shiny, dark red blood clot with a jelly-like consistency. After removing the blood clot, an AEF was identified, approximately 5 cm below the opening of the left subclavian artery. The fistula was 3 cm in length and was opened into the esophagus (Fig. 2). The edges of the AEF were smooth. Traces of blood were found in the esophagus, trachea, and bronchi. The stomach contained approximately 1,100 g of dark red, shiny blood clots with extensive fibrin stripes visible in the cross sections. Petechial hemorrhage was evident on the surfaces of the heart and lungs. Pallor of the internal organs was observed. Additionally, perimortem fractures of the left VII–VIII ribs at the parasternal line and the left III–VII ribs at the midclavicular line were present. Hepatic steatosis and stage II atherosclerotic changes in the coronary and cerebral arteries with lumen stenosis of up to 25% were also identified. No other significant changes in the internal organs were detected during the autopsy.

During the toxicology analysis, no ethyl alcohol was found in the blood, but its concentration in the urine was 0.33%.

Histological examination of the AEF border showed focal atrophy of the esophageal mucosa, hypertrophy of the muscular layer with focal fibrosis extending to the submucosa and to the junction with the aorta, mixed inflammatory infiltration, neovascularization, and focal necrosis (Fig. 3–5).



**Fig. 1.** A dissected, 16 cm long, 8 cm diameter aneurysm of the descending thoracic aorta

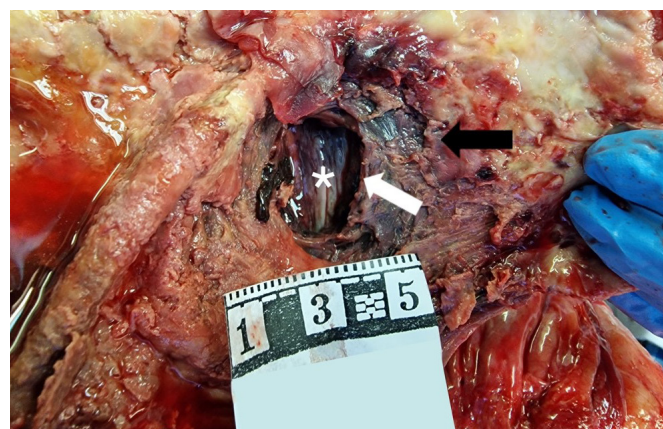


**Fig. 2.** Aortoesophageal fistula (white arrow) in the middle third of the aortic aneurysm (black arrow showing aorta) – 3 cm vertical wall defect, opening into the esophagus (white asterisk)

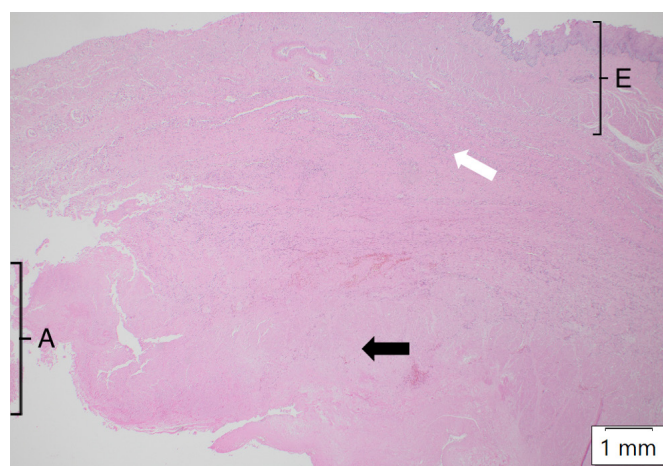


**Fig. 3.** Border of aortoesophageal fistula. A – aortic fragment. E – esophagus. White arrow – accumulation of inflammatory cells at different stages, histological structure is difficult to distinguish. Black arrow – necrotic debris, histological structure is disrupted. Hematoxylin and eosin (H&E),  $\times 20$

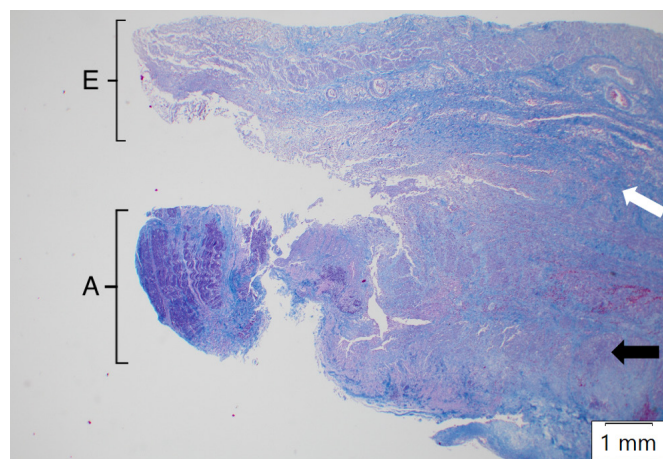


**Fig. 4.** Border of aortoesophageal fistula. A – aortic fragment. E – esophagus. White arrow – accumulation of inflammatory cells at different stages, histological structure is difficult to distinguish. Black arrow – necrotic debris, histological structure is disrupted. Masson's trichrome,  $\times 20$

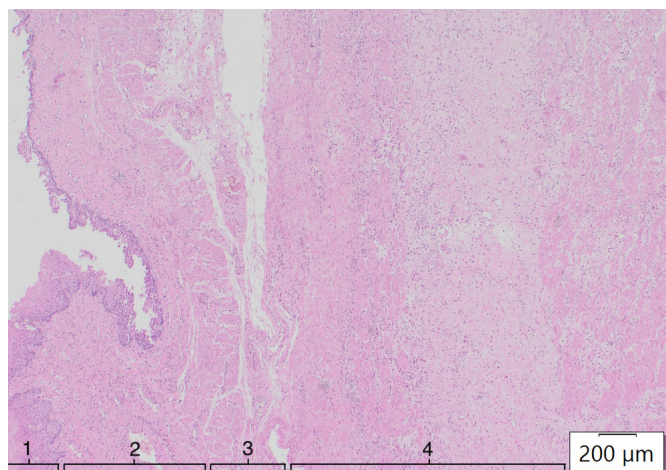


**Fig. 5.** Esophageal layers at the border of aorto-esophageal fistula: 1 – musoca, 2 – muscularis mucosae, 3 – submucosa, 4 – muscularis. H&E, ×40

## DISCUSSION

The described case highlights the sudden and fatal outcome of a patient with primary AEF and dissecting aneurysm of the descending thoracic aorta. The first case of AEF was described in the early 19th century [2, 12]. Nevertheless, AEF represents an exceedingly rare cause of upper gastrointestinal bleeding (UGIB) [3].

AEF forms due to primary or secondary causes [16]. A primary AEF develops spontaneously as a result of direct mechanical friction forces and inflammation within the aorta [16]. Primary AEF is usually located where the esophageal wall closely abuts the aortic arch [4, 17]. In this case, the AEF was found about 5 cm below the opening to the left subclavian artery. The main etiological factor of primary AEF, accounting for more than half of cases, is atherosclerotic aneurysm of the thoracic aorta [4, 18, 19]. The second most common cause of primary AEF, accounting for almost one-fifth of cases, is ingested foreign bodies [4]. Approximately 80% of foreign bodies are swallowed by children, especially under 3 years of age [4]. Esophageal cancer is another frequent cause of primary AEF, making up around 17% of cases [19–21]. AEF is less commonly caused by other aortic and esophageal diseases, including esophageal ulceration, gastroesophageal reflux disease, ingestion of corrosive substances, trauma, tuberculosis, and radiotherapy [19, 20]. Primary AEF occurs ten times less frequently than secondary AEF [22]. Aortic arch replacement, TEVAR to the descending aorta, and esophageal surgery are among the main causes of secondary AEF [23]. In recent years, the growing number of aortic surgeries and related procedures has increased the incidence of secondary AEF [3, 22].

In the presented case, autopsy data suggested that AEF formation was induced by a dissecting aneurysm of the descending thoracic aorta, 8 cm in diameter. According to the literature, thoracic aortic aneurysms with a diameter greater than 5 cm are at higher risk of dissection or rupture [24]. Pathogenetically, an aneurysm arises when the aortic wall weakens [25]. Loss of integrity in the blood vessel wall is predominantly due to atherosclerotic disease [25].

Pronounced aortic atherosclerosis was also identified in our presented case. High pulsatile intravascular pressure leads to gradual aneurysm enlargement [25]. A growing aortic aneurysm compresses the adjacent esophagus, causing local aseptic inflammation, necrosis, and the formation of adhesive granulation tissue between the aorta and the esophagus, resulting in AEF [4, 17, 25]. In this case, the pathological changes present in the walls of the aorta and the esophagus at the AEF site could be partly explained by the mechanisms described above.

In the described case, there was a lack of information regarding the symptoms in an individual's medical history. The subject was diagnosed with hypertensive heart disease, and a sclerotic aortic arch was incidentally discovered, both three years prior to his death. No evidence of treatment or follow-up was observed. Overall, diagnosing AEF is difficult and demands a high level of suspicion [21]. Vascular abnormalities tend to be overlooked because esophageal varices and peptic ulcer disease account for most UGIB etiologies [19]. In the case of AEF, bleeding is usually bright red, unlike in cases of bleeding from esophageal varices [4]. In this case, based on the findings of external and internal examination, the amount of blood loss was severe enough to cause death. Typically, AEF manifests as the Chiari's triad, which includes midthoracic pain or dysphagia, sentinel hemorrhage followed by an asymptomatic period, and massive arterial hematemesis [2, 4, 10, 11, 14, 15, 22, 25, 26]. All the symptoms of the Chiari's triad are present in only about 20–45% of cases [26]. Chest pain affects 59% of patients [17] and is provoked by esophageal leakage with mediastinitis, distension, or dissection of the aortic wall [4]. Sentinel hemorrhage occurs in 65% of cases [17]. Bleeding temporarily stops due to a sudden drop in blood pressure, which facilitates the formation of a blood clot [22]. In our presented case, it is likely that multiple bleeding episodes occurred, evident from the morphological features of blood clots inside the stomach, possibly indicating the sentinel hemorrhage event. Hours to days can pass between the sentinel hemorrhage and the final massive bleeding [22]. Other symptoms that may occur include back pain, fever, and sepsis [25].

Esophagogastroduodenoscopy (EGD), which is usually the initial diagnostic tool for UGIB, has limited diagnostic utility [21, 26]. In some cases of AEF, an EGD may reveal a pulsating mass, a bluish discoloration, or a blood clot in the esophagus [22]. However, the sensitivity of EGD for aorto-enteric fistula is only about 25% [26, 27]. In addition, there is a chance that clots will be dislodged during the EGD, leading to fatal bleeding [27]. To promptly confirm the diagnosis and initiate treatment, patients with suspected AEF should undergo computed tomography angiography [19], which reveals active blood extravasation from the aorta to the esophagus [22]. Unfortunately, in many cases, including the described case, the diagnosis of AEF is confirmed upon post-mortem examination [21].

AEF is usually a fatal condition [12]. The overall survival rate of AEF patients is low, and management is contentious

[21]. Nowadays, bleeding is usually controlled by performing TEVAR, which is less invasive, has a lower risk of complications, and is more suitable for hemodynamically unstable patients than surgery [15]. TEVAR has a high technical success rate of 87.3% [28]. After TEVAR, 15.2% of patients develop stent graft infection, and 13.8% of patients have recurrent AEF [28]. It is recommended to use TEVAR as a bridge therapy [28, 29]. Once the patient's condition has been stabilized, surgical procedures such as subtotal esophagectomy, mediastinal debridement, reconstruction of the aorta and the esophagus should be performed [28, 29]. Nevertheless, the optimal surgical strategies are still a matter of debate [28, 29].

## CONCLUSIONS

Aortoesophageal fistula is an infrequent and life-threatening phenomenon. Our presented case is an example of a primary aortoesophageal fistula with subsequent exsanguination as a complication of a dissecting aneurysm of the descending thoracic aorta. The cause of death was determined during the post-mortem examination. This study emphasizes the problematic nature of late diagnosis of aortoesophageal fistula. Autopsy plays an important role in determining the cause of death in such rare and clinically difficult-to-detect pathologies. In clinical practice, monitoring and managing thoracic aortic aneurysms is essential for reducing the risk of developing the aortoesophageal fistula.

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## Santrauka

### AORTOSTEMPLINĖ FISTULĖ – NUSILEIDŽIANČIOSIOS KRŪTININĖS AORTOS ANEURIZMOS KOMPLIKACIJA: KLINIKINIS ATVEJIS

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**Įvadas.** Aortostemplinė fistulė yra reta būklė, apibrėžiama kaip patologinė jungtis tarp aortos ir stemplės. Dėl didelės kraujavimo iš viršutinės virškinamojo trakto dalies rizikos ši fistulė yra gyvybei pavojinga, todėl pacientų išgyvenamumas yra mažas. Nors Chiari triada laikoma klasikine šios būklės išraiška, ji pasireiškia mažiau nei pusė aortostemplinės fistulės atvejų, o galutinė diagnozė dažnai patvirtinama po mirties.

**Klinikinio atvejo aprašymas.** Savo bute rastas negyvas 65 metų vyras. Įvykio vietoje ant vyro veido ir grindų, šalia kūno, buvo matomos kraujo dėmės. Autopsijos metu nustatyta aortos aterosklerozė, nusileidžiančiosios krūtininės aortos disekuoji aneurizma ir 3 cm vertikalios aortostemplinės fistulės, esančios 5 cm žemiau kairiosios poraktinės arterijos atsišakojimo. Skrandyje rasta kraujo krešulių (1100 g) su fibrino sankaupomis, kas leidžia įtarti ankstesnį kraujavimo epizodą.

**Diskusija ir išvados.** Aortostemplinė fistulė yra gyvybei grėsmę kelianti būklė. Aprašytas klinikinis atvejis atskleidžia šios fistulės ankstyvos diagnostikos iššūkius ir autopsijos svarbą nustatant mirties priežastį retų patologijų atveju. Siekiant sumažinti aortostemplinės fistulės išsivystymo riziką, klinikinėje praktikoje svarbu optimali pacientų, turinčių aortos aneurizmą, stebėseną ir gydymą.

**Reikšminiai žodžiai:** aortos aneurizma, aortostemplinė fistulė, autopsija, kraujavimas, teismo medicina.

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