

*Prikaz slučaja /
Case report*

PRIMARY AORTO-ENTERIC FISTULA:
Case report

PRIMARNA AORTOENTERIČNA FISTULA:
Prikaz slučaja

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Abstract

Aorto-enteric fistula is a rare pathological entity of communication between the lumen of the aorta and the cavity of the digestive tract. In the non-treatment cases, mortality is almost 100%, and in the operative treatment cases, mortality remains high, up to 70%. The aim of our work was to describe one case of the patient with primary aorto-enteric fistula. In conclusion, aorto-enteric fistula should be suspected in cases of gastrointestinal bleeding and the presence of an aortic aneurysm. Early diagnosis of this pathological entity posed by an overview of computerized tomography and proper treatment are crucial.

INTRODUCTION

Aorto-enteric fistula (AEF) is a rare pathological entity of communication between the lumen of the aorta and the cavity of the digestive tract, described at the beginning of the 19th century that complicate the fatal gastrointestinal bleeding (1-3). In the non-treatment cases, mortality is almost 100%, and in the operative treatment cases, mortality remains high, up to 70%.

The aim of our work was to describe one case of the patient with AEF and to search and display relevant literature on this topic.

Case Report

The 70-year-old male patient was admitted to the Emergency Center of Military Medical Academy due to blood vomiting and blood stools several times on the day of admission. He says he had two episodes of blood vomiting over the past week, which spontaneously stopped. During the admission, in the department, the patient threw up about 300 ml of fresh blood. The patient complains of epigastric pain. The overview shows blood pressure of 100/60 mmHg, pulse 90 beats/min, palpated pain sensitivity in the epigastrium. The patient states that 14 years ago he has operation for abdominal aortic aneurysm (AAA). Laboratory findings:

white blood cells $19.10 \cdot 10^9/L$; red blood cells $2.92 \cdot 10^{12}/L$; haemoglobin 94.2 g/L; haematocrit 29%; and platelets $212 \cdot 10^9/L$. The patient was referred to an emergency multislice computed tomography (MSCT) examination of the abdomen.

An overview was made on the 64-slices Toshiba MSCT per protocol for the MSCT examination of abdomen. The existence of suprarenal AAA, 9.5 cm in a diameter, with mural thrombus and circulating lumen was determined. The aneurism is in close contact and compresses to the back wall of the D3 and D4 duodenal segment, with signs of extravasation of the contrast medium from the lumen of the aorta to the lumen of the duodenum (Figures 1-3). There is no clear demarcation between the anterior wall of the aneurysm and the posterior wall of the duodenum. There are no signs of extralumination of the contrast medium intra- and retroperitoneal. Based on MSCT findings, diagnosis of AEF was established and the patient was admitted for emergency surgery.

The AEF operation was performed by a vascular and general-abdominal surgeon. Intraoperative condition was established after an earlier resection of infrarenal aneurysm, with the interposition of tubular synthetic graft, the existence of visceral patterns and the changed anatomical rela-

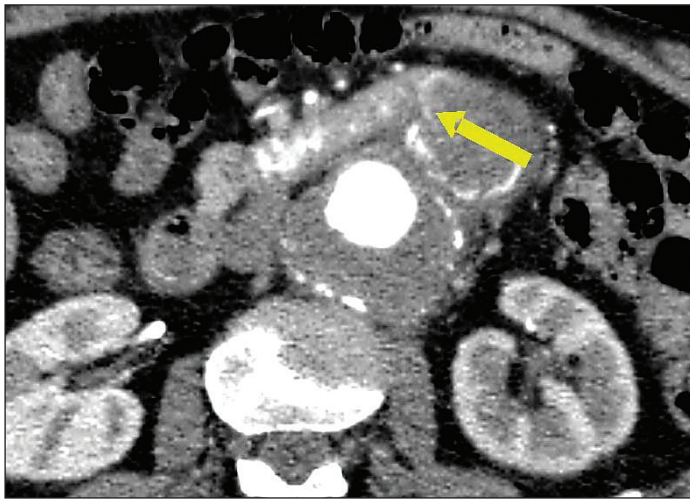


Figure 1. Axial MSCT: Aortoenteric fistula (arrow)

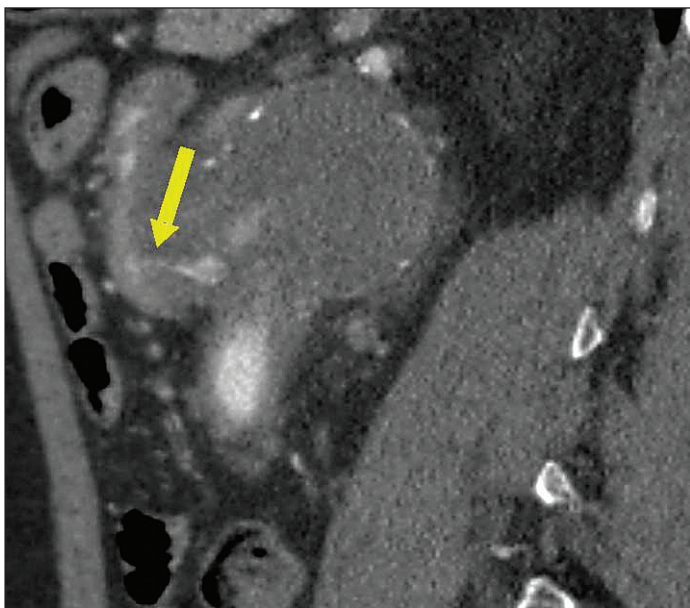


Figure 2. Sagittal MSCT: Aortoenteric fistula (arrow)

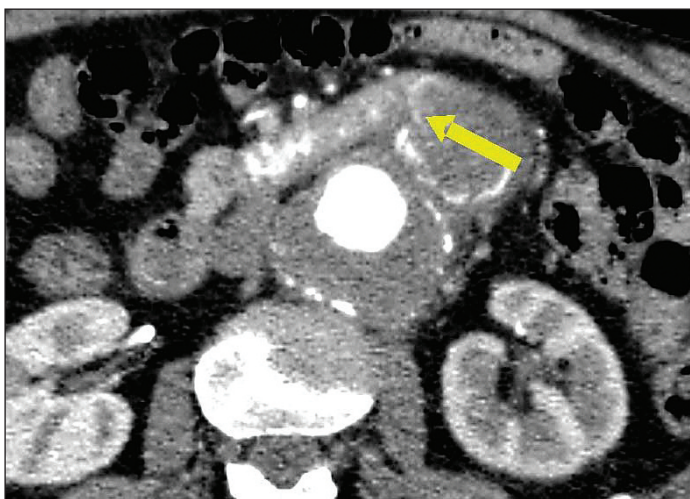


Figure 3. Axial MSCT: Blood in small bowel (arrows)

tionships of retroperitoneal structures. Cranial from proximal anastomosis, AAA altered diameter of 9 cm, with mural thrombus. The front wall adheres to the rear wall of the D4 segment of the duodenum in the presence of AEF diameter

of 3 mm. In addition to the duodenal duct, partial resection of the suprarenal portion of the abdominal aorta (AA) was performed. Distal and proximal patches are blindly closed, while subcutaneous bifurcation by-pass is established in order to form axilobifemoral anastomosis.

DISCUSSION

This paper presents the case of the patient with primary AEF who was diagnosed with MSCT after several episodes of blood vomiting. This was about AEF between the AA and the rear wall of the D4 segment of the duodenum.

The AEF is a pathological communication between the lumen of the aorta and the cavity of the digestive tract, most often of the small intestine. About 80% of cases involve the communication of the aorta and the D3 or D4 segment of the duodenum (4). In relation to etiology there are primary and secondary AEFs. Primary de novo, most often on the terrain of direct contact with aneurysmal enlarged AA (most often infrarenal part of the aorta) and part of the digestive tract, most often D3 of the duodenum (5, 6). Experimental research supports the thesis that primary AEFs are primarily direct contact, that is, the mechanical force, and then the infection, the main etiopathogenesis factors (7). They are very rare with an incidence of 0.007 per million (8, 9), respectively an incidence rate of 0.1-0.8% of all AEF (6). The AEFs are associated with a high mortality rate (65-100%). Secondary AEF can occur from two weeks to ten years after aortic surgery, i.e. placement of aortic stent graft, with an incidence of up to 2% (10, 11) and occur on the terrain of the aorta graft infection, and therefore the infectious agent is the main etiological factor (12).

The most common cause of primary AEF is atherosclerotic aortic aneurysm. Other conditions that may be the causes of AEF include tuberculosis, syphilis, infection, cancer, pre-therapeutic radiation, and foreign bodies (13, 14). The true cause of bleeding is often misdiagnosed as an ulcer or other condition and therefore no adequate measures are taken to prevent long and often fatal bleeding. The reasons for this repeated bleeding are not entirely clear, but are probably due to the spasm of the duodenal muscle layer in response to bleeding and hypotension due to hypovolemia (6).

This is a difficult, urgent condition that is most often manifested in the so-called "Herald bleeding" (3, 15), i.e. episodes of gastrointestinal transient or self-limiting bleeding accompanied by abdominal pain, which may precede hours or weeks of "catastrophic" bleeding. In addition to gastrointestinal bleeding, typical symptoms are pulsated abdominal mass and abdominal pain, but this triage of symptoms meets in 23% of patients with AEF (2). In 70% of patients, different forms of bleeding occur: hematemesis, hematochezia, melena and chronic anemia.

In patients who do not require prompt surgical care, the diagnostic method of choice is computed tomography (CT) (16) with variable sensitivity and specificity (17), although it has been shown earlier in the studies that the sensitivity is about 94% and the specificity is about 85% in the detection of the AEF scanner. Direct signs indicating the

existence of AEF are the finding of the extravasation of the contrast agent from the aorta to the lumen of the digestive tract and/or of the free gas around the aorta or in its lumen, as well as focal bowel-wall thickening, disruption of the aortic fat cover (9, 13). In addition to CT diagnosis, it can also be placed in aortography, magnetic resonance imaging, ultrasonography, and esophago-gastro-duodenoscopy (18).

Primary AEFs are treated with en bloc resection and graft reconstruction, extra-anatomical bypass, and endovascular stenting (14, 19). Due to massive and deadly bleeding, without surgery, primary AEF causes safe death, which is why early detection and appropriate surgical intervention are important. In cases of severe, repeated bleeding, the only reasonable measure that can be applied is surgery. Therefore, there is a principle that whenever there is gastrointestinal bleeding and aortic aneurysm, immediate surgical interventions should be undertaken and the possible existence of AEF (17) should be resolved. In the largest pub-

lished series so far to 118 cases of aortoduodenal fistula, only 25% of patients were treated with surgery, and 60% survived the operation (20).

Today, in addition to traditional AEF treatment with open surgery, it is also tried with endovascular procedures as an alternative therapeutic option (2, 3, 21). An endovascular approach is currently recommended as a transient procedure for unstable patients, followed by definitive open surgery, if possible.

CONCLUSION

Primary AEF is a rare and fatal disease. Therefore, AEF should be suspected in cases of gastrointestinal bleeding and the presence of an aortic aneurysm. Early diagnosis by MSCT examination and proper treatment are of indispensable significance.

Sažetak

Aortoenterična fistula jeste redak patološki entitet komunikacije između lumena aorte i šupljine digestivnog trakta. U slučaju ne tretiranja smrtnost je skoro 100%, a u slučaju operativnog tretmana smrtnost je i dalje visoka, do 70%. Cilj našeg rada je bio da se opiše jedan slučaj pacijenta sa primarnom aortoenteričnom fistulom. Zaključeno je da treba uvek postaviti sumnju na aortoenteričnu fistulu u slučajevima gastrointestinalnog krvarenja i postojanje aneurizme aorte. Rana dijagnoza ovog patološkog entiteta postavljena pregledom kompjuterizovanom tomografijom i pravilni tretman su od presudnog značaja.

REFERENCES

- Cooper A. Lectures on the Principles and Practice of Surgery. London: Astley Cooper; 1829.
- Spanos K, Kouvelos G, Karathanos C, Matsagkas M, Giannoukas AD. Current status of endovascular treatment of aortoenteric fistula. *Semin Vasc Surg.* 2017;30(2-3):80-4.
- Varetto G, Gibello L, Trevisan A, Castagno C, Garneri P, Rispoli P. Primary Aortoenteric Fistula of a Saccular Aneurysm: Case Study and Literature Review. *Korean Circ J.* 2015;45(4):337-9.
- Bernard VM. Aortoenteric fistulas. In: Rutherford RB, editor. *Vascular surgery*. 3rd ed. Philadelphia, Pa: Saunders; 1989: 528-35.
- Tareen AH, Schroeder TV. Primary aortoenteric fistula: two new case reports and a review of 44 previously reported cases. *Eur J Vasc Endovasc Surg.* 1996;12(1):5-10.
- Costea R, Vasiliu EC, Zărnescu NO, Neagu S. Primary aortoenteric fistula: case report. *Chirurgia (Bucur).* 2015;110(1):78-80.
- Ikonomopoulos DC, Spanos PK, Lazarides DP. Pathogenesis of aortoenteric fistula. An experimental study. *Int Angiol.* 1986;5(1):33-7.
- Voorhoeve R, Moll FL, de Letter JA, Bast TJ, Wester JP, Slee PH. Primary aortoenteric fistula: report of eight new cases and review of the literature. *Ann Vasc Surg.* 1996;10(1):40-8.
- Hughes FM, Kavanagh D, Barry M, Owens A, MacErlaine DP, Malone DE. Aortoenteric fistula: a diagnostic dilemma. *Abdom Imaging.* 2007;32(3):398-402.
- Reilly LM, Altman H, Lusby RJ, Kersh RA, Ehrenfeld WK, Stoney RJ. Late results following surgical management of vascular graft infection. *J Vasc Surg.* 1984;1(1):36-44.
- O'Hara PJ, Hertzner NR, Beven EG, Krajewski LP. Surgical management of infected abdominal aortic grafts: review of a 25-year experience. *J Vasc Surg.* 1986;3(5):725-31.
- Busuttill RW, Rees W, Baker JD, Wilson SE. Pathogenesis of aortoduodenal fistula: experimental and clinical correlates. *Surgery.* 1979;85(1):1-13.
- Lee CW, Chung SW, Song S, Bae MJ, Huh U, Kim JH. Double primary aortoenteric fistulae: a case report of two simultaneous primary aortoenteric fistulae in one patient. *Korean J Thorac Cardiovasc Surg.* 2012;45(5):330-3.
- Sever A, Rheinboldt M. Unstable abdominal aortic aneurysms: a review of MDCT imaging features. *Emerg Radiol.* 2016;23(2):187-96.
- Malik MU, Ucbilek E, Sherwal AS. Critical gastrointestinal bleed due to secondary aortoenteric fistula. *J Community Hosp Intern Med Perspect.* 2015;5(6):29677. doi: 10.3402/jchimp.v5.29677.
- Mark AS, Moss AA, McCarthy S, McCowin M. CT of aortoenteric fistulas. *Invest Radiol.* 1985;20(3):272-5.
- Wood A, Bendjelid SM, Bendjelid K. Primary aortoenteric fistula: should enhanced computed tomography be considered in the diagnostic work-up? *Anesth Analg.* 2005;101(4):1157-9, table of contents.
- Perrone T, Pagani C, Mossolani EE. Ultrasound detection of aortoenteric fistula in a patient with sepsis. *J Ultrasound.* 2017;20(2):157-9.
- Vilas-Boas F, Azevedo F, Marques M, Baldaque-Silva F, Cardoso H, Costa-Lima J, et al. Primary aortoenteric fistula. *Clin J Gastroenterol.* 2013;6(4):299-302.
- Sweeney MS, Gadacz TR. Primary aortoduodenal fistula: manifestation, diagnosis, and treatment. *Surgery.* 1984;96(3):492-7.
- Rossi UG, Cariati M. Aortoenteric fistula. *J Cardiovasc Comput Tomogr.* 2015;9(5):461-2.

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