

# Aortoenteric fistulas — case report series from the Department of Surgery of the 4<sup>th</sup> Military Clinical Hospital in Wrocław over the years 2010–2016

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## Abstract

**Introduction.** Aortoenteric fistulas constitute a rare, but severe clinical problem involving a pathological connection between the abdominal aorta and gastrointestinal tract. We distinguish primary and, much more frequent, secondary fistulas. All such fistulas require urgent surgical treatment. Currently, the most common treatment method involves open in situ prosthesis replacement or an extra-anatomical by-pass. Endovascular procedures offer an alternative to open surgery, but remain controversial.

**Material and methods.** The analysis included 6 surgical procedures in 5 patients with aorto-duodenal fistula treated at the Department of Surgery of the 4. Military Clinical Hospital over the years 2010–2016. Open surgery was performed in 4 cases, while 2 patients underwent stentgraft implantation. Diagnostics consisted of an angio-CT and upper gastrointestinal tract endoscopy. All patients were diagnosed with radiological signs of prosthesis infection, 2 of 6 patients died during the perioperative period. Surgical complications were observed in 5 of 6 cases (83%).

**Results.** Endovascular treatment was associated with a reduction in the number of early postoperative complications and shortening of hospital stay.

**Conclusions.** Based on the results and literature data, stentgraft implantation into the aorto-duodenal fistula appears to be a viable alternative treatment method.

**Key words:** stentgraft, aorto-duodenal fistula, aortic prosthesis, abdominal aorta, gastrointestinal hemorrhage

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## Introduction

Aortoenteric fistula is a rare condition that involves a pathological connection between the aorta and the

gastrointestinal tract. In about 80% of cases, fistulas are formed between the third and fourth section of the duodenum and the adjacent abdominal aorta. In 15% of cases, the connection develops between aorta

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and small intestine [1–4], and in 5% between aorta and large intestine. We may distinguish primary aortoenteric fistulas (PAEF) and secondary aortoenteric fistulas (SAEF).

PAEF is a rare complication associated mainly with an abdominal aortic aneurysm and advanced atherosclerosis. PAEF is identified in 0.04–0.07% of autopsies [5]. Typical symptoms include gastrointestinal bleeding, pulsatile abdominal mass and abdominal pain.

SAEF is more common than PAEF, with its incidence reaching 0.77–1.6% in patients after prosthesis implantation due to abdominal aortic aneurysm. It is thought that the main cause of fistula formation involves mechanical irritation of the posterior wall of duodenum by a layer of sutures in the upper anastomosis of the prosthesis and subsequent inflammatory process in the adjacent tissues [1, 5–7]. SAEF may be suspected in patients after abdominal aortic aneurysm surgery presenting with recurrent gastrointestinal bleeding and signs of systemic infection [1, 2, 6, 7].

Untreated aortoenteric fistulas are associated with 100% mortality. Quick diagnosis and appropriate treatment give the patient a chance to survive, although treatment is associated with a high risk of complications and perioperative death, reaching 19–77% depending on the surgical method [6, 8, 9]. To date, open surgery has played a major role in the treatment of aortoenteric fistulas, although endovascular treatment is being used in an increasing number of patients.

## Material and methods

Five patients with aortoenteric fistulas have been admitted and treated at the Department of Surgery of the 4. Military Clinical Hospital over the years 2010–2016. One patient underwent surgery twice due to recurrence of the aortoenteric fistula. All cases involved secondary fistulas in patients who had undergone prosthesis implantation due to abdominal aortic aneurysm. All fistulas developed between abdominal aorta and the 3<sup>rd</sup> or 4<sup>th</sup> section of the duodenum. All patients were male, aged 59–73 years (mean age 66.8 years). The time between implantation of the vascular prosthesis and the occurrence of symptoms ranged from 5 months to 9 years (mean 41.8 months). Patients were acutely admitted to hospital and every one underwent computed angiotomography. Gastroscopy was performed in 5 cases. In total, 6 repair surgeries were performed: 4 primary *in situ* prosthesis replacement procedures (silver-coated prosthesis) with primary duodenal closure, and 2 stentgraft implantations. In one case, endovascular prosthesis implantation was

accompanied by the application of retroperitoneal drainage and microjejunostomy formation with gastric drainage using Taylor's method.

## Results

All patients were acutely admitted to the Department. Upper gastrointestinal bleeding (5 of 6 patients — 83%) was the most frequent complaint that prompted hospital admission. Fever > 38°C was present in 4 cases (66%) and 2 patients reported abdominal pain (33%). Laboratory findings included leukocytosis ranging from  $13.5 \times 10^3/\text{mm}^3$  to  $31.7 \times 10^3/\text{mm}^3$  (mean  $15.4 \times 10^3/\text{mm}^3$ ) and elevated C-reactive protein (CRP) level ranging from 43 mg/L to 139 mg/L (mean 61 mg/L).

Infection of endovascular prosthesis was described in all angio-CT studies, although fistula was visualized in only 4 patients (66%). The dominating radiological symptoms included widening of periaortic tissues due to inflammation (100%), inflammatory infiltration of the duodenum (4 cases, 66%), and presence of periaortic gas bubbles (3 patients, 50%).

Endoscopy was performed in all patients with symptoms of gastrointestinal bleeding. The source of bleeding was found in 2 cases (40%), but attempts at stopping the hemorrhage failed. In one patient examination revealed a thrombus at the site of the fistula without active bleeding.

Following the examinations and diagnosis of SAEF, all patients were referred for urgent surgery. In four cases the infected vascular prosthesis was removed and replaced with a silver-coated prosthesis. In all cases, duodenal repair was performed during the same surgery.

Two patients who had been treated with classic open surgery died: one patient due to massive intraoperative hemorrhage and the second patient because of multiorgan failure in the course of systemic infection 34 days after surgery. Infection of the surgical wound was noted in two patients and one patient was reoperated due to acute ischemia of the right leg. In one case recurrence of aorto-duodenal fistula was diagnosed, accompanied by massive hemorrhage and retroperitoneal abscess. Mean hospitalization time among patients treated with open surgery amounted to 37 days. One patient was considered fully recovered after a 24-month follow-up period.

Microbiological examination of material obtained from the area of aorto-duodenal fistula revealed G(+) bacteria, i.e. *Staphylococcus aureus* MRSA in 2 patients and *Staphylococcus epidermidis* in another 2 cases. Blood cultures demonstrated systemic infection in all patients infected with the above pathogens. Prolonged antibiotic

**Table 1.** Patients undergoing surgery due to secondary aortoenteric fistulas

Patient	Age	Time from surgery (months)
1	62	28
2	59	61
3	71	36
4	73	11
5	69	110
5*	69	5

\*Patient 5 was reoperated due to fistula recurrence

therapy with vancomycin was applied according to the antibiogram. A G-negative *Pseudomonas aeruginosa* was identified in 1 patient who died during surgery.

Two patients were treated with endovascular stent-graft implantation (EVAR) technique. In one case the procedure was performed in a patient with recurrent aorto-duodenal fistula and massive gastrointestinal bleeding. Additionally, angio-CT study revealed a retroperitoneal abscess. On the second day after surgery, following stabilization of patient's general condition, microjejunostomy was performed and retroperitoneal drainage was maintained for 14 days. At the same time, gastric drainage using Taylor's method was applied in order to decompress the gastrointestinal tract. In the second case, the procedure consisted of stentgraft implantation followed by therapy with intravenous and oral antibiotics. Patient treated with EVAR due to recurrent fistula required reoperation due to pseudoaneurysm in the area of inferior anastomosis of aortobifemoral prosthesis. Serial angio-CT studies performed 6, 12, and 18 months after the procedure did not reveal inflammatory lesions in the periaortic space. The second patient was readmitted to hospital 8 months later due to fever and leukocytosis. Computed angiotomography showed persistent inflammatory changes in the periaortic region. Follow-up examination performed after implementation of intravenous antimicrobial agents and 6 months of oral antibiotics demonstrated regression of inflammatory lesions, the absence of systemic symptoms and normalization of leukocyte levels. Mean hospitalization time among patients treated with EVAR amounted to 29 days.

## Discussion

Aortoenteric fistulas are rare, late complications of aortic prosthesis implantation. The third and fourth section of the duodenum is the most frequent site of fistula formation (80%), followed by small intestine (15%) and colon (5%) [1–4, 9, 11].

**Table 2.** Comparison of complication rates for open surgery/ endovascular aortic repair (OS/EVAR) in our material

Complication	OS	EVAR
Death	2	–
Wound infection	2	1
Acute limb ischemia	1	–
Sepsis	1	1
Recurrent fistula	1	–
Periaortic abscess	1	–

In 71% of cases, SAEF is secondary to implantation of aortic prosthesis due to the abdominal aortic aneurysm and in 38% after implantation of aortobifemoral prosthesis in the course of Leriche syndrome [8, 10]. The exact pathogenesis of fistula formation has not been entirely elucidated, although more frequent occurrence of fistulas following surgical repair of aortic aneurysm implies an association with earlier weakening of duodenal wall and greater aortic diameter [1, 11, 12]. Typical symptoms demonstrated by patients with SAEF include coffee-ground vomit and melena. They are observed in 60–76% of cases and constitute the most common symptom [1–3, 8, 10, 13]. In our material gastrointestinal bleeding was observed in 5 of 6 cases (83%). However, bleeding site or thrombus was identified in only 3 patients (50%). Visualization of the source of bleeding may be impaired, as fistulas are usually located in the 3<sup>rd</sup> and 4<sup>th</sup> section of the duodenum; thus, it is only successful in 40–76% of cases [14–16]. Gastrointestinal hemorrhage is often accompanied by general symptoms of infection (fever, leukocytosis, elevated CRP), which are present in 30–87% of cases [1, 14, 17, 18]. In the presented material 3 of 6 patients (50%) presented with symptoms of sepsis and 2 of 6 patients (33%) reported abdominal pain. Less frequent symptoms accompanying SAEF include pulsatile abdominal mass, lower limb ischemia, prosthesis occlusion, and back pain [14, 19].

Computed angiotomography is the primary and the most reliable diagnostic modality. Its sensitivity is estimated at 94% and specificity reaches 85% [14, 20, 21]. In our material, inflammation of the periaortic space associated with prosthesis infection was identified in all patients. However, direct visualization of aorto-duodenal fistula was only possible in 4 of 6 studies (66%). Ultrasonography and angiography may play an adjuvant role in the diagnosis of SAEF [8, 19]. Untreated SAEF is associated with almost 100% mortality and for that reason, surgical intervention should be implemented as quickly as possible [4]. Urgent treatment is associated with 60% mortality, decreasing to 38% if the patient may be prepared for the procedure [14]. Current therapeutic options include open surgery (OS) or endovascular procedure (EVAR). Open surgery usually involves

removal of the infected aortic prosthesis and replacing it with silver-coated prosthesis [1, 4, 8, 11, 14]. Alternatively, rifampicin-soaked prosthesis, homograft, or patient's own vein may be used [6, 10]. Extra-anatomical axillofemoral bypass is another treatment option [1, 10, 14, 18]. Open surgery is associated with a very high risk of complications. Thirty-day mortality after the procedure reaches 30–56% [1, 8, 11, 14]. Postoperative complications occur in up to 77% of patients treated with an open method [11, 14, 20]. In the presented material 5 procedures were performed urgently and one after prior preparation. Mortality reached 50% (2 of 4 patients). One death was associated with massive intraoperative hemorrhage, the other with multiorgan failure in the course of septic shock. Complications occurred in 2 of 3 (66%) operated subjects who survived the surgery.

Endovascular treatment involves implantation of a stentgraft to cover the fistula. EVAR may be used as ad hoc or definitive treatment. In the first case, the aim of stentgraft implantation is to control bleeding and stabilize the patient's condition. Subsequently, after preparing the patient for surgery and implementing long-term antibiotic therapy, open in situ prosthesis implantation or axilofemoral bypass grafting is performed [1, 6, 11, 14]. The time period between EVAR and OS ranges from 300 to 567 days [6, 11]. In the case of definitive treatment, stentgraft implantation is the final procedure. Comparing EVAR to OS one may note significant reduction in rates of early mortality (0% vs. 35%) and other complications (25% vs. 77%) [14, 20]. Meantime of hospital stay is also shortened — 19.4 vs. 44 days [21]. However, EVAR is associated with significantly greater risk of sepsis. During the two years after the procedure, the risk of sepsis after EVAR amounts to 42%, while for OS it is 19% [1, 11]. The risk of developing sepsis after EVAR during the first two years may be achieved through the application of antibiotic therapy for a period of at least 6 months. With one antibiotic the risk amounts to 63%, in the absence of antibiotic therapy — 100% [1, 11, 20]. Lifelong use of a single antibiotic should be considered after EVAR [11]. There were no deaths after EVAR in our material. One patient developed an inguinal pseudoaneurysm that required repeat surgery. One patient required hospitalization 8 months after the procedure due to the symptoms of sepsis, which resolved after prolonged antibiotic therapy. One EVAR procedure, performed in order to control massive gastrointestinal bleeding in a patient with recurrent SAEF, required laparotomy due to the presence of a periaortic abscess.

## Conclusions

SAEF is a rare, severe complication of aortic prosthesis implantation. It usually involves the 3<sup>rd</sup> and 4<sup>th</sup> section of the duodenum.

Treatment of SAEF is associated with high perioperative mortality and high risk of complications.

Each patient after gastrointestinal hemorrhage with a history of aortic prosthesis implantation should undergo diagnostics toward SAEF.

EVAR remains controversial as a treatment option for SAEF due to the fact that stentgraft is implanted into an infected region. However, it significantly reduces mortality and complication rates.

EVAR may be used both as an ad hoc life-saving procedure supplemented by delayed open surgery, as well as a definitive treatment option.

Long-term antibiotic therapy in order to reduce the risk of sepsis as a late complication plays an important role in all patients treated for SAEF.

Open surgery seems to be the method of choice for patients who may be prepared for surgery and with predicted long survival time. Endovascular procedures are better suited for treatment of massive hemorrhages and for patients in poor general condition.

## Conflict of interest

None.

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