

JAN GNUS^{1, 2, A, B, D}, STANISŁAW FERENC^{1, A, B, D}, MAGDALENA KOŚCIELNA^{3, B, C},
MAŁGORZATA PAPROCKA-BOROWICZ^{1, 2, C}, PIOTR DAWIDCZYK^{1, C},
MAŁGORZATA DZIEWISZEK^{1, B}, WOJCIECH WITKIEWICZ^{1, E, F}

Secondary Aortoenteric Fistula After Abdominal Aortic Graft Implementation in Our Own Material

¹ Department of General and Vascular Surgery, Voivodship Specialist Hospital in Wrocław, Poland

² Department of Clinical Biomechanics and Physiotherapy in Motor System Disorders, Wrocław Medical University, Poland

³ Research and Development Centre, Voivodship Specialist Hospital, Wrocław, Poland

A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
D – writing the article; E – critical revision of the article; F – final approval of article

Abstract

Background. Secondary aortoenteric fistula is a rare but severe complication occurring after abdominal aortic graft implementation.

Objectives. The aim of our study was to review the clinical presentation, diagnosis and postoperative course of patients with aortoenteric fistulas following vascular operations on the abdominal aorta in the years 2000–2014.

Material and Methods. Among all the patients treated in our center, during a 10-year period, aortoenteric fistulas were observed in 24 cases. The first symptoms occurred between 4 weeks and 8 years after the procedure (3.4 years on average). The most common clinical presentation was gastrointestinal bleeding in 16 cases (66.7%). All patients underwent surgical repair of an aortoenteric fistula, with graft removal and replacement *in situ* using silver impregnated prosthesis.

Results. The 30-day mortality rate after surgical treatment of SAEF was 37.5% (9 patients). Causes of death included: hemorrhagic shock, multi-organ failure and myocardial infarction. The early complications after the surgical repair of an aortoenteric fistula occurred in 19 (79.2%) patients. Mortality during the one-year follow-up period was 38.5% (5 patients) and the one-year complication rate was 69.2%.

Conclusions. Each patient with a history of abdominal aortic graft implementation and presenting symptoms of gastrointestinal bleeding requires careful diagnosing for aortoenteric fistula. New methods of surgical treatment offering lower mortality should be developed (*Adv Clin Exp Med* 2016, 25, 6, 1265–1271).

Key words: complications, aortic surgery, GI-bleeding, aortic diseases, aortoenteric fistula.

A secondary aortoenteric fistula (SAEF) occurs after abdominal aortic graft implementation due to an aneurysm or atherosclerosis in 0.4–2.4% of cases [1–3]. Several factors are involved in the pathogenesis of SAEF: graft infection, bowel erosion caused by anastomotic pseudoaneurysm, contact of the pulsatile aortic graft with the duodenum leading to erosion of the graft through the bowel wall, and possible injury to the bowel during graft implementation [4–7]. The most common clinical presentation of SAEF is severe gastrointesti-

nal bleeding, often preceded by mild, self-limiting “herald bleeding”. Other symptoms include: abdominal pain, back pain, hemorrhagic shock, septic shock, or a combination of hemorrhage and sepsis [5, 8–10]. A variety of diagnostic procedures are involved in the diagnosis of SAEF, e.g., contrast enhanced CT and gastroduodenoscopy [11, 12]. However, diagnosis can be made using an ultrasound, and sometimes an initial diagnosis is based only on a physical examination and the history of aortic graft placement. Patients with SAEF

require surgery, which is the only definitive treatment, but associated with a high mortality rate of between 18.8–59% [13–15]. The aim of our study was to review the clinical presentation, diagnosis and postoperative course of patients with aortoenteric fistulas following vascular operations on the abdominal aorta in the years 2000–2014.

Material and Methods

In the years 2000–2014, on the Vascular Ward, there were 1081 aortic prostheses implemented: 368 women (34%) and 713 men (66%) aged between 42 and 86 (58.8 on average). With the help of a computer database, 24 patients (6 women and 18 men) diagnosed with aortoenteric fistulas and treated in our hospital in the aforementioned period were identified. Only patients with a history of operations of any type regarding the abdominal aorta were taken into account. We analyzed the medical documentation regarding patients' hospitalization due to the primary procedure and hospitalization due to a secondary aortoenteric fistula. In the analysis, information regarding the type and the cause of the primary operation, complications following the operation, clinical presentation of patients diagnosed with SAEF, diagnostic procedures performed, postoperative course, surgical site culture, blood culture and antimicrobial treatment were taken into consideration. All patients discharged from the hospital following SAEF repair surgery were followed up in the Vascular Surgery Outpatient Clinic in our hospital. The follow-up visits were scheduled after 2, 6 and 12 months in the first year after the surgery, and every 12 months in the following few years and included a physical examination, laboratory tests and Duplex Doppler ultrasound imaging. If any abnormalities were detected, abdominal angiographic CT examination was carried out and, if necessary, patients were hospitalized for diagnosis and further treatment. The endpoint of the study was either the success or failure of the treatment of patients diagnosed with aortoenteric fistula. The treatment was successful in cases where the patients showed no symptoms of graft infection, fistula recurrence or aortic graft thrombosis during the follow-up.

Results

Indications for the primary procedure included: aortoiliac occlusive disease in 13 (54.1%) cases, abdominal aortic aneurysm (AAA) elective repair in 9 (37.5%) cases, and AAA emergency repair in 2 (8.3%) cases. Of those 24 patients, complica-

tions occurred in 6 cases during the initial procedure, among which there were 4 cases of proximal anastomotic bleeding, 1 case of *vena cava* reconstruction and 1 case of renal vein reconstruction following iatrogenic injury. Complications during the 30-day period after the initial procedure were noted in 9 (37.5%) patients and included 4 cases of fever over 38.5°C. Two patients were re-operated due to graft limb thrombosis, and 3 patients underwent relaparotomy: 2 patients due to retroperitoneal bleeding and 1 patient due to colon ischemia.

Among the 24 patients with aortoenteric fistulas, 16 patients presented symptoms of gastrointestinal bleeding: hematemesis or melena, 6 hemorrhagic shock, 4 septic shock, 8 abdominal pain, 4 back pain and 8 pulsatile abdominal mass (Table 1). The symptoms among those 24 patients occurred between 4 weeks and 8 years after the primary procedure (3.5 years on average). Gastrointestinal bleeding symptoms had a recurrent character in 4 patients. While diagnosing those patients the following examinations were taken into consideration: medical history, clinical examination, contrast enhanced CT, gastroduodenoscopy, colonoscopy, angiography, and abdominal ultrasound. On the basis of the clinical examination and medical history, 6 patients who presented with shock underwent immediate surgery. Among all of those patients, the presence of aortoenteric fistula was confirmed during the surgery. The other 18 patients were operated on, on average 4 days after admission to the ward (between 12 h and 15 days). Among diagnostics procedures, 14 patients underwent abdominal ultrasound (35.7% accuracy) 12 gastroduodenoscopy (33.3% accuracy), 10 contrast enhanced CT (70% accuracy), 4 angiography (50% accuracy), and 8 patients were examined *via*

Table 1. Symptoms of aortoenteric fistulas among 24 patients with the implemented vascular prosthesis in the years 2000–2014

Symptoms	Number of patients
Gastrointestinal bleeding	16
Hematemesis	7
Melena	9
Shock	10
hemorrhagic	6
septic shock	4
Pain	12
abdominal	8
back	4
Pulsating abdominal mass	8

*Each patient may have more than 1 symptom.

Table 2. The diagnosis of patients with aortoenteric fistulas treated in the years 2000–2014

Type of examination	Number of patients	Positive diagnosis	Accuracy (%)	95% CI
USG	14	5	35.7	0.5086 to 0.8855
Gastroduodenoscopy	12	4	33.3	0.5003 to 0.9029
CT	10	7	70	0.3595 to 0.7844
Angiography	4	2	50	0.2957 to 0.9075
Colonoscopy	8	0	0	0.6278 to 1.0000

colonoscopy (0% accuracy) (Table 2). In 22 patients the fistula was located in the duodenum; the jejunum was involved in 2 (8%) cases.

In all the patients the abdominal aorta was reached *via* the transperitoneal approach, and the graft was removed and replaced *in situ* with a silver impregnated prosthesis. The bowel wound was closed with a double layer of sutures and additionally covered with the adjacent omentum. Intraoperative periaortic tissue and graft material cultures were obtained. Blood cultures were taken from all patients who survived the surgical procedure. The overall 30-day mortality rate was 37.5% (9 patients), 2 patients died intraoperatively from hemorrhagic shock. The main cause of 30-day mortality was postoperative hemorrhagic shock due to massive blood loss (3 patients). Other causes included multi-organ failure, myocardial infarction and hemorrhagic shock due to rupture of the proximal anastomosis (Table 3). Intraoperative cultures of 20 patients were evaluated positive. Among the positive cultures of periaortic tissues and graft material, there were 12 only Gram (+) or Gram (–) species including *Staphylococcus* (3 cases of MRSA), *Streptococcus* spp. and *Escherichia coli*, while 8 contained mixed flora: *Pseudomonas aeruginosa*, *Enterococcus* spp., *Veilonella* spp., *Peptostreptococcus* spp., *E. coli*, and *Staphylococcus aureus* (Table 4). Positive postoperative blood cultures were obtained from 6 patients (Table 5). In the preoperative period all patients received empiric antimicrobial treatment which included the intravenous administration of cefuroxime plus metronidazole. Antibiotic therapy was continued postoperatively for at least one week, and in the case of positive blood cultures was prolonged up to 1.5 months and modified, if necessary, based on sensitivities from blood culture. Antibiotic therapy regimen was based on recommendations of our local microbiologist. In-hospital complications after surgical treatment for aortoenteric fistula occurred among 19 patients. Seven patients once again underwent operations: thrombectomy due to lower limb ischemia was performed in 2 cases. Five patients underwent relap-

Table 3. Perioperative mortality of patients with aortoenteric fistula

Causes of death	n	%
Postoperative hemorrhagic shock	3	12.5
Intraoperative hemorrhagic shock	2	8.3
Multi-organ failure	2	8.3
Rupture of proximal anastomosis	1	4.2
Myocardial infarction	1	4.2

Table 4. Type of flora isolated from the fragments obtained from 20 patients with aortoenteric fistula

Type of flora	Type of pathogen	Number of patients	%
Single	<i>Staphylococcus</i> spp.	6 (3 MRSA)	50
	<i>Streptococcus</i> spp.	4	33.3
	<i>E. coli</i>	2	16.6
Mixed	<i>Staphylococcus aureus</i>	5	62.5
	<i>E. coli</i>	4	50
	<i>Enterococcus</i> spp.	3	37.5
	<i>Pseudomonas aeruginosa</i>	3	37.5
	<i>Veilonella</i> spp.	1	12.5
	<i>Peptostreptococcus</i> spp.	1	12.5

Table 5. Type of flora isolated from positive blood cultures

Pathogen	n
MRSA	3
<i>E. coli</i>	2
<i>Enterococcus faecalis</i>	1

arotomy: 2 patients due to intestinal obstruction, 2 patients because of retroperitoneal bleeding from the proximal anastomosis, and retroperitoneal abscess in 1 patient. In-hospital complications are list-

Table 6. Complications after aortoenteric fistula surgery in the years 2000–2014

Complications	n	%
In-hospital complications		
cardiac complications		
(arrhythmia/MI)	4	18.2
persistent sepsis	3	13.6
lower limb ischemia	2	9.1
intestinal obstruction	2	9.1
retroperitoneal bleeding	2	9.1
renal insufficiency	2	9.1
pneumonia	1	4.5
exenteration	1	4.5
retroperitoneal abscess	1	4.5
Overall	19/22	86.3
Late complications		
graft infection	4	26.7
lower limb ischemia	2	13.3
recurrent fistula	1	6.6
abdominal wall abscess	1	6.6
femoral pseudoaneurysm	1	6.6
Overall	9/15	60.0

* Each patient may have more than one complication.

ed in Table 6. Nine (37.5%) patients died. The most common cause of postoperative death was hemorrhagic shock due to massive blood loss despite massive transfusion of blood and plasma. Other causes of death are listed in Table 3. Late complications occurred in 9 cases (Table 6). The most common late complication was graft infection in 4 patients. Two patients with graft infections had to undergo re-surgery. Both re-operated patients died within 5 days after surgery. In one of these patients, a recurrent aortoduodenal fistula was diagnosed 7 months after the primary surgery due to SAEF. In the remaining 2 patients, symptoms of infections were resolved following antibiotic therapy. During the follow-up, 5 patients died. Two (13.3%) patients died due to late complications following SAEF repair surgery. The cause of death in both patients was multi-organ failure due to sepsis resulting from aortic graft reinfection. In the remaining 3 patients (20%), death was caused by cardiovascular complications: myocardial infarction and congestive heart failure. The treatment was successful in 41.7% of cases (10/24 patients).

Discussion

Secondary aortoenteric fistula was described for the first time by Brock in 1953, two years after the first abdominal aortic homograft implementation due to aneurysm. The description concerned

a fistula located between the homograft and duodenum [16]. An aortoenteric fistula between the intestine and the aortic prosthesis was first described in 1956 (Claytor et al.) [17]. In 1958, McKenzie et al. demonstrated the first successful repair of a secondary aortoenteric fistula between a synthetic graft and the intestine [18]. Secondary aortoenteric fistula is a rare, but potentially fatal late complication following the implementation of an aortic prosthesis. According to some authors, SAEFs are more common following abdominal aortic aneurysm surgeries. In the study by Armstrong et al. [19], in 71% of patients with SAEF the primary procedure was abdominal aortic aneurysm repair. According to Bergqvist et al., aortoiliac occlusion accounted for 38.5% of the cases (128/332), while abdominal aortic aneurysm surgery accounted for 43.1% of the cases (143/332). Among the cases in our study, the most common primary procedure was Y-graft implantation due to aortoiliac occlusion. Similar results were reported by Kuestner et al. [20]. Owing to its non-specific clinical presentation, it is difficult to diagnose SAEF correctly in a sufficiently short period of time. Gastrointestinal bleeding was the most common symptom in our patients; hematemesis or melena occurred in 16 patients (66.7%). Other symptoms included: abdominal pain (8 patients), back pain (4 patients), and a pulsating abdominal mass found in 8 patients during the physical examination. Similar data was reported by, e.g., Sierra et al. [21]. In our own experience supported by literature data, patients with symptoms of gastrointestinal bleeding and a history of aortic graft implantation should undergo a particularly careful diagnostic examination for SAEF. Of note, however, are also the reports of SAEF cases not associated with aortic grafts. In their recent work, Luo et al. [22] reported the case of such a fistula between the rectum and the left common iliac artery following an aortoiliac aneurysmorrhaphy procedure. In the same paper, the authors presented a review of the literature available in English regarding the incidence of SAEF not associated with aortic grafts. They listed 22 cases of SAEFs not associated with grafts, following such procedures as endarterectomy in the aortoiliac section, aortic suture due to trauma, and aneurysmorrhaphy. Bearing these reports in mind, one should suspect SAEF in patients with gastrointestinal bleeding following any procedure regarding the aorta. Moreover, SAEFs are not always manifested by massive gastrointestinal bleeding. According to Leon Jr. et al. [23], 430 cases of aortoenteric erosion were reported, in which the outer graft surface was in communication with the lumen of the gastrointestinal tract without graft continuity being impaired.

In these cases the bleeding, if any, is caused by damage to the mucosal membrane by the pulsating graft. In such patients SAEF may be manifested only by septic conditions of varying intensity. The following diagnostic procedures were involved in the diagnosis of SAEF in our patients: gastroduodenoscopy, abdominal contrast enhanced CT, abdominal ultrasound, angiography and colonoscopy. The highest accuracy in diagnosing SAEF on our ward was achieved by abdominal contrast-enhanced CT (70%). The accuracy of other diagnostic procedures was 35.7% (abdominal USG) and 33.3% (gastroduodenoscopy). Angiography was performed in 4 patients and SAEF was diagnosed in 2 cases. Even contrast enhanced CT, which is the most accurate procedure (79% accuracy according to Bergqvist et al.) [13], rarely allows for imaging of the aortoenteric fistula. The diagnostic procedure was focused on indirect radiology symptoms which, with reference to the clinical presentation of a patient, most frequently may suggest the presence of an aortoenteric fistula: air in periaortic tissues later than 4 weeks following aortic surgery, focal thinning of the intestinal wall, absence of tissue between the aortic wall and the intestine, liquid in periaortic tissues or their swelling later than 3 months following aortic surgery. These symptoms may also suggest infection of the aortic prosthesis or fistula occurrence. Preliminary diagnosis of SAEF was made if the above-mentioned symptoms co-existed with gastrointestinal bleeding. The relatively low accuracy of gastroduodenoscopy results from the fact that the operator was not always able to obtain an image of the distal duodenum part, where fistulas are located most frequently, or the image was masked by fresh blood or a massive thrombus. The total inefficiency of colonoscopy in diagnosing SAEF in our patients was associated with the fact that there were no cases of secondary fistulas with colon involvement. There have been, however, numerous cases reported worldwide in which a colonoscopy facilitated a definite diagnosis. Such cases were published by, e.g., Malaki et al. and Perez et al. [24, 25]. Bergqvist et al. reported 25 published cases of SAEFs with colon involvement, which accounted for 8% of all reviewed cases. A colonoscopy should not be excluded from the panel of diagnostic tests to be performed in patients suspected of having SAEF. Apart from allowing definite diagnosis to be made, colonoscopy is also useful in ruling out other causes of gastrointestinal bleeding in patients following aortic surgery. The average time between admission to hospital and diagnosis and surgery in our patients was 4 days (from 12 h to 15 days) except for 6 patients who had emergency surgery owing to hemorrhagic shock following admission to hospi-

tal. Similar data was reported by Armstrong et al. [19]. Recognized surgical treatment methods in SAEF include graft removal and axillobifemoral bypass, graft removal and new graft *in situ*, suture of the bowel and patch plasty of the aorta. In our center we used graft removal and a new, silver-impregnated graft *in situ*, a method that was used by, for example, Kavanagh et al. [26] and Batt et al. [27]. Other revascularization methods include, e.g., allograft implantation or femoral vein application [28–30]. However, preparatory procedures (a femoral vein) or preparation (a blood group compatible allograft) are not always possible when the patient with bleeding caused by an aortoenteric fistula has urgent surgery [31]. The use of silver-impregnated graft shortens the operation time and, currently, is a widely accepted technique. The perioperative mortality rate in our patients was 37.5% and was similar to the rate reported by Bergqvist et al. [13] (38–47%) referring to the type of surgery we performed. In another systematic review, Aguilar et al. reported even lower early mortality rate of 8–13.3% [32]. The perioperative mortality rates for the most popular method globally (graft removal and axillobifemoral bypass) reported by the same authors were 42–51% [13] and 18.2–44% [32], respectively. Infection factors play a significant role in the pathogenesis of SAEF. Intraoperative cultures of 20 patients were evaluated positive. Gram-positive *cocci* were most frequently isolated bacteria. Gram-negative strains most frequently from mixed flora which also included *Enterococcus* spp., *Pseudomonas aeruginosa* and anaerobic bacteria (*Veilonella* and *Peptostreptococcus*). In all patients, preoperative empiric antimicrobial treatment with cefuroxim or ciprofloxacin and metronidazole was introduced. The antibiotic policy was introduced in our hospital during the study period. Antimicrobials were divided into Tyree classes. Without consultation with the clinical microbiologist, physicians are allowed to provide empirical therapy using drugs belonging to the first class, i.e. amoxicillin and clavulonate or ciprofloxacin with or without metronidazole. Most strains isolated from SAEF patients included gram-positive *cocci*, including multiresistant strains. Therefore, administration of glucopeptide in combination with fluoroquinolone should be considered in the empirical therapy of these patients. In patients with positive blood cultures, a postoperative treatment based on antibiogram was introduced for at least a 2-month period. Similar data regarding antimicrobial treatment was reported by Saers et al. [33]. The incidence of reinfection appears to be a significant limitation of the *in situ* graft replacement method. In our patients, the rate of reinfection amounted to 26.7%. Compared to the report by Batt et al., who

employed a similar treatment method, this rate was relatively high. However, it must be borne in mind that in the report published by Batt et al., SAEF patients comprised only 37.5% of all treated patients. We would also like to point out that only 2 (13.3%) patients treated in our hospital required reoperation. We believe that *in situ* implantation of silver-impregnated grafts is the optimal solution due to reduced intraoperative trauma, relatively low perioperative mortality and acceptable reinfection rates. The fact is, however, that due to the rare occurrence of aortoenteric fistulas, no gold standard in surgical treatment methods or perioperative management have yet been developed. Currently, it is considered that the stent graft is the source of infection, so it is recommended to remove the graft and, if possible, implant the prosthesis [34]. Axillobifemoral bypass and graft re-

moval is associated with the relatively lowest mortality rate (31%) according to Bergqvist et al. [13], but such a surgical procedure is only possible in hemodynamically stable patients. Deshpande et al. [35] described a promising method of endovascular SAEF repair in which the patient is hemodynamically stabilized to allow surgeons time for the implementation of an extra-anatomic bypass and correct fistula repair.

A secondary aortoenteric fistula is a rare but severe postoperative complication following the implementation of an aortic prosthesis. Every patient with symptoms like gastrointestinal bleeding and one who has a previously implemented aortic prosthesis requires careful examination for SAEF. Owing to the high mortality rate of SAEF repairs, new methods for surgical treatment offering lower mortality rates should be developed.

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Address for correspondence:

Małgorzata Dziewiszek
Department of General and Vascular Surgery
Voivodship Specialist Hospital in Wrocław
ul. Kamieńskiego 73A
51-124 Wrocław
Poland
E-mail: mdziewiszek@gmail.com

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