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AORTODUODENAL FISTULA THREE YEARS AFTER AORTOBIFEMORAL BYPASS: CASE REPORT AND LITERATURE REVIEW

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SUMMARY – Secondary aortoenteric fistulas (SAEF) are a relatively rare but dangerous complication of aortal reconstructive surgery. We present a patient that underwent aortobifemoral bypass three years before developing the signs of aortoenteric fistula, and we reviewed the literature on the topic. Since the clinical signs are nonspecific, physicians should have a high index of suspicion for SAEF in patients who underwent aortal reconstructive surgery. The most useful diagnostic tools for stable patients are upper gastrointestinal endoscopy and computed tomography scan with contrast that can, in combination with history and clinical signs, enable accurate diagnosis in more than 90% of patients. Unstable patients with suspected aortoenteric fistula should undergo exploratory laparotomy. The treatment of choice is open surgery with graft excision, wide debridement of infected tissue, bowel repair or resection followed by an extra-anatomic bypass or *in situ* placement of a new graft. Early postoperative mortality remains high, around 30% in most analyses. Currently there are no guidelines for the diagnosis and management of SAEF, so individualized approach is necessary for each patient.

Key words: *Aortoenteric fistula; Upper gastrointestinal bleeding; Postoperative complications; Aortobifemoral bypass; Endovascular procedures*

Introduction

Gastrointestinal (GIT) bleeding is one of the common conditions in emergency department. The most common causes are peptic and duodenal ulcers, tumors and esophageal varicosities. A rare but clinically important cause of upper GIT bleeding is aortoeneric fistula (AEF), which is an abnormal communication between the aorta and the intestine. Spontaneously acquired communication is classified as primary AEF, originating mostly from abdominal aneurysm communicating with the bowel, while secondary aortoeneric fistula (SAEF) is a result of medical manipulation (complication of aortic repair after stent/prosthesis or graft placement)[1,2].

This report presents a case of aortoduodenal fistula 3 years after an uncomplicated revascularization procedure of external iliac artery by aortobifemoral bypass grafting, with review of the literature.

Case Report

A 54-year-old man was admitted to our hospital with ileus, hematochezia and hemodynamic instability in October 2009. In 2006, he underwent Y-prosthesis placement (aortobifemoral bypass), and in 2007 he was admitted for observation due to hematochezia. Diagnostic work-up revealed large bowel polyposis and sigmoid diverticulosis by endoscopic procedures. A month before admission, angiography was per-

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formed due to intermittent claudication and at that time, the findings were normal.

On admission, blood tests revealed normocytic anemia (hemoglobin 119 g/L) and elevated infection parameters (leukocytes 15x10^9/L, C-reactive protein 58 mg/L). Upper GI endoscopy showed blood and coagula in the duodenum with no evident origin of blood loss. Computed tomography (CT) scan revealed ectopic air around the Y-prosthesis (Fig. 1). Due to massive hemorrhage, urgent explorative laparotomy was performed, which revealed an aortoduodenal fistula, which connected the 3rd segment of the duodenum and the proximal part of the Y-prosthesis. The fistula was probably the result of local infection and direct mechanical erosion of the duodenal wall by the graft itself. The connection between the duodenum and the prosthesis was removed first and a patch plastic was performed (excision of the duodenal defect with primary closure). Thereafter, Silver Dacron graft was used to replace the initial Y-prosthesis. Greater omentum was interposed between the intestine and the aortic wall. Ileostomy followed due to ileus and for intestinal decompression. The patient was discharged 14 days after the surgery in a relatively good condition.

The patient was readmitted in December 2009 because of deep vein thrombosis and bleeding from the rectum. CT angiography showed contrast leakage into the retroperitoneum (Fig. 2). Emergency exploratory laparotomy revealed adhesions between the aorta and the jejunum and proximal graft dehiscence with retroperitoneal hematoma, which penetrated to sigmoid colon or rectum; the exact point of penetration was not found. The tubular and the left arm of the prosthesis were removed because of graft infection. The right arm was not infected, thus it was left *in situ*. Aortoiliac continuity was restored by a left axillofemoral bypass (6 mm PTFE bypass, left with silver graft). The rest of the postoperative period was complicated with left axillofemoral bypass thrombosis, which eventually led to above-knee amputation of the left leg.

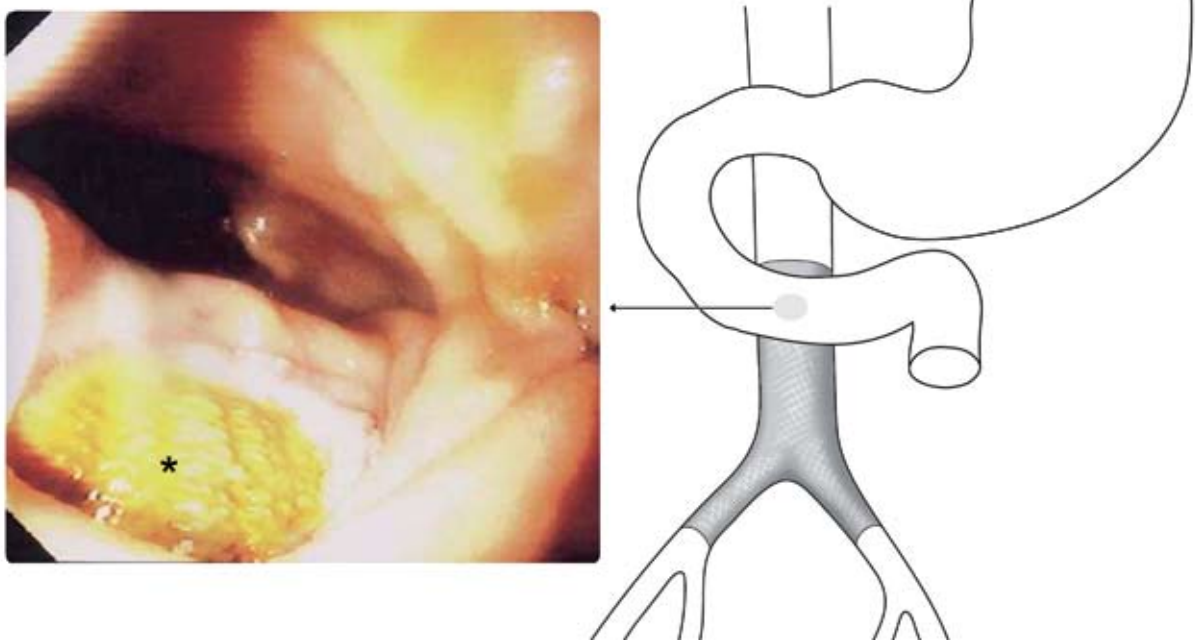
Fig. 1. Transverse abdominal CT scan with contrast showing perigraft gas (arrow).

Fig. 2. Sagittal abdominal CT scan with contrast showing contrast leakage into the retroperitoneum.

Discussion and Literature Review

Secondary AEF is a possible complication of aortic reconstruction with prosthetic implants. An annual incidence of 0.6%-2% has been reported for secondary AEFs[3,4]. The most common site of SAEF formation is in the 3rd and 4th portion of the duodenum (80% of cases), with other locations (esophagus, small and large bowel) being possible but rare[5,6]. The interval between the surgery and clinical manifestation ranges

Fig. 3. Diagram of graft-enteric erosion (type 2) and upper GI endoscopic snapshot of graft (*) erosion into the lumen of the duodenum without blood leakage (seen as yellow area and asterix).



from several days to several years (range, 2 days to 27 years)[7,8].

The etiology of SAEF is multifactorial and not well understood. Two types of SAEF according to the etiology and clinical signs have been described.

Type 1, a graft-enteric fistula (GEF), is a communication between the bowel and the disrupted graft-aorta anastomotic site. The cause is disruption of the anastomotic line (due to, e.g., perigraft infection, errors in technique, deterioration of the vessel wall graft dilatation) and subsequent leakage. This can then lead to anastomotic pseudoaneurysm, perigraft hematoma and erosion into the bowel. Most GEFs occur at the proximal aortic anastomosis and involve distal duodenum. This is the most common type of fistula (around 75% of all cases). The main clinical sign is upper GI bleeding in the form of either hematemesis or melena, and can be massive causing quick exsanguinations[9]. We assume that this type of secondary AEFwas present in our case at the first urgent operation.

Type 2, a graft-enteric erosion (GEE) or a paraprosthetic-enteric fistula (Fig. 3), is a necrosis of the enteric wall overlying the graft caused by mechanical pressure of the graft to the bowel. This results in bathing of the graft by enteric contents and chronic graft infection without direct involvement of the suture line. GEE accounts for around 20% of SAEF. The most common symptoms are related to recurrent bacteremia (low-grade fever, malaise, weight loss) and occur in 75% of patients. Mild or moderate GI bleeding occurs in around 20% of patients. Other symptoms including abdominal pain, septic emboli in lower extremities, septic arthritis, multicentric osteomyelitis, and hypertrophic osteoarthropathy have been described[9,13]. Considering this, deep vein thrombosis in our patient could be caused by septic emboli originating from paraprosthetic-enteric fistula.

SAEF is difficult to diagnose due to its variable clinical manifestations, which results in high mortality. A review of 18 studies indicated a median time to correct diagnosis of eight days (range, 0 hours to 18 months)[10]. In many cases, the reason for delayed diagnosis is mostly due to the fact that the signs of SAEF are nonspecific (Table 1)[11]. The SAEF development

Table 1. Prevalence of clinical signs in patients with secondary aortoenteric fistula (modified from Pipinos et al.*[11])*

Clinical sign	Prevalence (%)
Gastrointestinal bleeding	88
Sepsis	44
Abdominal pain	30
Back pain	15
Groin mass	12
Abdominal pulsatile mass	6

can be subtle, at first the 'herald' bleeding is detected, which is then followed by massive bleeding within hours or months after the first bleeding[12].

To date, no diagnostic method has been shown to have the sensitivity and specificity to be considered a criterion standard for the diagnosis of SAEF. If the patient presents with GI bleeding, the first-line diagnostic method is usually esophagogastroduodenoscopy, which is essential in identifying and evaluating upper GI bleeding. Nevertheless, it has been shown that it detects AEF in less than 50% of cases[13,14]. In most cases, CT scan with contrast will enable the diagnosis of SAEF when combined with history data. Two analyses that included 55 and 22 patients showed that CT scan had a sensitivity of up to 94% and specificity up to 85% for diagnosing perigraft infection with or without AEF, and if GI bleeding was present, the diagnosis was made in 100% of cases[15,16]. After aortic surgery, CT scan normally shows perigraft soft tissue edema, fluid, and ectopic gas. However, after 3-4 weeks, any ectopic gas is abnormal and should be considered a sign of perigraft infection and possibly fistulization to bowel, which should resolve in 2-3 months postoperatively[15]. Other CT findings that are suggestive of both perigraft infection and AEF include pseudoaneurysm, loss of normal fat plane between the aorta and the adjacent bowel, and disruption of the aortic wall. Because of the overlapping CT features, SAEF and perigraft infection may be difficult to differentiate. This is where positron emission tomography (PET) can be used because the method is highly sensitive for infection processes[17]. When combining PET and CT in one session, it is possible to distinguish infective processes on the prosthesis from the processes around it[18]. However, the findings that strongly correlate with the presence of SAEF include ectopic gas, focal bowel wall thickening, breach of the aortic wall, and extravasation of contrast material into the bowel lumen[15,16,19,20].

Although conventional angiography is often requested for evaluation of GI bleeding, extravasation of aortic contrast into the adjacent intestinal lumen that is diagnostic of AEF is rarely seen and therefore is this method not routinely used[20]. If the patient presents with massive bleeding and is unstable, emergency exploration is indicated.

Treatment options for SAEF include open surgery and endovascular treatment. The classic surgical treatment includes graft excision, wide debridement of infected tissue beds (if present), bowel repair or resection followed by graft explantation alone or *in situ* replacement with autologous vein, allograft or prosthetic material or extra-anatomic revascularization and graft excision. *In situ* autologous graft replacement seems to be the best choice to treat young patients with longer life expectancy, whereas *in situ* prosthetic graft replacement is better choice in emergencies and older patients. Furthermore, the type of intestinal closure is important. It is suggested that smaller defects should be closed with direct sutures, while larger ones should be managed with resection and reanastomosis. Also, the extension of graft removal has been discussed. The results have shown that limited resection does not correlate with graft reinfection[21,22]. Broad-spectrum antibiotics should be given. To prevent complications, atraumatic operative technique, aseptic operative field, parenteral antibiotics and proper covering of the graft with periaortic tissue (e.g., omentum) are important. Some authors perform a temporary extra-anatomic bypass before excision of the prosthesis to prevent long-term ischemia in already high-risk patients. Mortality after open surgery (extra-anatomic bypass or *in situ* graft replacement) during early postoperative period remains high; it was around 30% in several retrospective analyses, rising to around 50% after several years of follow-up (range, 2-5 years)[23,24]. No significant differences in graft patency and rate of reinfection were observed when comparing the two open surgical procedures. On the other hand, newer studies proved the importance of graft material. They observed that *in situ* rifampin-soaked grafts or silver-grafts with omental wrapping and long-term anti-

biotics were associated with a lower rate of reinfection[25,26].

Endovascular treatment has been considered for SAEF management in larger centers. Endovascular treatment was associated with a lower mortality rate, but on the other hand, reinfection and recurrent bleeding were reported in 44% of patients[27]. That is why endovascular treatment is a bridge operation before the open procedure in patients that are unstable or have many comorbidities[28-30].

Due to the poor outcome of SAEF, more attention should be paid to its prevention. The performance of end-to-end anastomosis with a short stump and high bifurcation enables the graft position in the plane of aorta and contact reduction with the intestine. Moreover, graft coverage with free or attached omentum reduces the contact between the intestine and the prosthesis[31-33]. Also, the role of suture material has been associated with SAEF formation[34].

Conclusion

SAEF are relatively rare but dangerous complications of aortal reconstructive surgery. Since the clinical signs are nonspecific, physicians should have a high index of suspicion of SAEF in patients that underwent aortal reconstructive surgery. The most useful diagnostic tools for stable patients are upper GI endoscopy and CT scan with contrast. The treatment of choice is open surgery with graft excision, wide debridement of infected tissue, bowel repair or resection, followed by an extra-anatomic bypass or *in situ* placement of a new graft. Early postoperative mortality remains high, around 30% in most analyses. Currently, there are no guidelines for the diagnosis and management of SAEF, so individualized approach is necessary for each patient.

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Sažetak

AORTODUODENALNA FISTULA TRI GODINE NAKON AORTO-BIFEMORALNE PREMOSNICE: PRIKAZ SLUČAJA I PREGLED LITERATURE

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Sekundarne aorto-enteralne fistule su rijetka ali opasna komplikacija aortne rekonstruktivne kirurgije. Prikazuje se bolesnik kod kojega su se razvili simptomi aorto-enteralne fistule tri godine nakon ugradnje aorto-bifemoralne premosnice, uz pregled literature. S obzirom na to da su klinički znaci vrlo nespecifični kod ovih bolesnika, važno je rano posumnjati na moguć razvoj sekundarne aorto-enteralne fistule kod bolesnika koji su bili podvrgnuti aortnoj rekonstruktivnoj kirurgiji. Od dijagnostičkih metoda najkorisnije su ezofagogastroduodenoskopija i kompjutorizirana tomografija s kontrastom koji, u kombinaciji s anamnezom i kliničkim znacima, omogućavaju postavljanje dijagnoze u preko 90% bolesnika. U nestabilnih bolesnika kod kojih postoji sumnja na razvoj ovoga stanja indicirana je eksplorativna laparotomija. U liječenju metoda izbora je laparotomija, ekscizija proteze, debrideman upaljenog tkiva, popravak defekta na crijevu ili resekcija zahvaćenog segmenta, te ekstraanatomska premosnica ili postavljanje nove proteze. Poslijeoperacijska smrtnost je oko 30%. Kako zasad nema smjernica za dijagnostiku i liječenje bolesnika sa sekundarnim aorto-enteralnim fistulama nužan je visoko individualizirani pristup za svakog bolesnika.

Ključne riječi: *Aortoenteralna fistula; Gornji probavni sustav, krvarenje; Poslijeoperacijske komplikacije; Aorto-bifemoralna premosnica; Endovaskularni postupci*