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Denise MD Özdemir-van Brunschot, Giel G Koning, J Adam van der Vliet

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Case Report: A 77-year-old Caucasian male who underwent a conventional repair of an inflammatory infrarenal aortic aneurysm, was readmitted with hematemesis only six weeks after surgery. Gastroscopy and computed tomography angiography indicated an aortoduodenal fistula and urgent aortic reconstruction was performed. An aortoduodenal fistula is a potentially lethal complication after transperitoneal open repair of an abdominal aortic aneurysm.

Conclusion: An aortoduodenal fistula seldom occurs as early after conventional transperitoneal open aneurysm repair as in our case. The early occurrence of the aortoduodenal fistula can be explained by the inflammatory character of the aneurysm.

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CASE REPORT

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Introduction: An aortoduodenal fistula is a potentially lethal complication after transperitoneal open repair of an abdominal aortic aneurysm. **Case Report:** A 77-year-old Caucasian male who underwent a conventional repair of an inflammatory infrarenal aortic aneurysm, was readmitted with hematemesis only six weeks after surgery. Gastroscopy and computed tomography angiography indicated an aortoduodenal fistula and urgent aortic reconstruction was performed. An aortoduodenal fistula is a potentially lethal complication after transperitoneal open repair of an abdominal aortic aneurysm. **Conclusion:** An aortoduodenal fistula seldom occurs as early after conventional transperitoneal open aneurysm repair as in our case. The early occurrence of the aortoduodenal fistula can be explained by the inflammatory character of the aneurysm.

Keywords: Aortoenteric, Aortoduodenal fistula, Conventional abdominal aortic aneurysm repair, Abdominal, Aneurysm, Inflammatory, Nevelsteen procedure

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INTRODUCTION

An aortoduodenal fistula was first described by Sir Astley Cooper in 1829 and is defined as an abnormal communication between the aorta and duodenum [1]. Communications of the aorta with other sites of the gastrointestinal tract, such as jejunum, stomach and sigmoid are possible, but are seen less often [2].

CASE REPORT

A 77-year-old Caucasian male patient was admitted to the emergency department because of general discomfort, nausea and dark stained emesis. Medical history of the patient included diabetes mellitus type 2, a peritonitis after appendicitis and peripheral artery occlusive disease (PAOD) stage 2a. Six weeks earlier, he underwent a conventional transperitoneal repair of an inflammatory infrarenal aortic aneurysm measuring 7.6 cm in diameter. It was a difficult procedure because of dense adhesions between the duodenum and the aortic aneurysm wall, leading to a small serosal injury which was sutured peroperatively. Six days after surgery the patient was discharged after an uneventful postoperative recovery.

At readmission, the vital signs were stable and hemoglobin level was 6.1 g/dL. Shortly thereafter, the patient produced hematemesis. A gastroscopy was performed subsequently. This examination revealed a vulnerable, edematous mucous membrane with a trace of blood in the duodenum. A computed tomography

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angiography (CTA) was performed (Figure 1) because of the high suspicion of an aortoduodenal fistula. Although no clear contrast extravasation in the duodenum was seen on this scan, an aortoduodenal fistula was presumed because of the very close proximity of the duodenum to the aorta and the air bubbles in and around the aneurysm sac and the signs of local bowel wall thickening.

An urgent aortic reconstruction was performed with a graft constructed from the right superficial femoral vein according to Nevelsteen (Figure 2) [3]. Intraoperatively, no signs of a duodenal perforation were detected. An additional femoro-femoral crossover bypass was constructed using a Dacron prosthesis (8 mm Gelsoft) because of absent pulsations in the left leg.

Perioperative cultures of the removed aortic graft turned out positive for *Streptococcus anginosus* and *Haemolytic streptococcus group B*.

The fourth day after surgery, a sudden resuscitation setting occurred and hemoglobin level declined to 3.5 g/dL. An emergency CTA was done, which showed no active blush. Despite all measures to resuscitate his condition worsened. Aortic stump blow-out was considered, but since no active blush was seen and it was considered unlikely. After careful discussion and consideration among all treating physicians and his family, it was agreed not to initiate any further surgical nor endovascular intervention and the patient deceased shortly afterwards. Permission for a postmortem examination was declined by the relatives.

DISCUSSION

Aortoduodenal fistulas are difficult to diagnose because patients often have non-specific complaints such as general discomfort, weakness and weight loss and imaging is seldom clear [2, 4]. Classical signs are gastrointestinal bleeding, a pulsatile abdominal mass and abdominal pain. Gastrointestinal bleeding can present as melena (up to 50%) and/or hematemesis (up to two-thirds). Typically, this bleeding is extensive but may be preceded by intermittent bleeding or herald bleeding. The sensitivity and specificity of computed tomography ranges from 40–90% and from 33–100%, respectively [4]. Signs include perigraft gas or fluid, soft tissue inflammation with edema, loss of continuous wrap of tissue around the graft and bowel wall thickening. Characteristics of other modalities, such as magnetic resonance imaging/angiography (MRI/MRA), have not yet been sufficiently evaluated [4].

A secondary aortoduodenal fistula is fatal without surgical intervention [3]. Treatment should consist of resection of the aortoduodenal fistula and preferably also the aortic graft. Revascularization can be undertaken in a variety of ways, including primary aortic repair, aortic replacement with a new prosthetic or venous graft or an extra-anatomical bypass (for example, axillo-bifemoral bypass) [4]. Each procedure is associated with

Figure 1: Computed tomography angiography (arterial phase) showing gas bubbles (arrow) in the aneurysm sac and a very close (adhesive) relation with duodenum.

Figure 2: Intraoperative view autologous venous aortic reconstruction (Nevelsteen procedure).

significant complications including lower extremity amputations, aortic stump blow out and mortality [3, 5]. Like conventional aortic repair, recently more reports are published of patients treated with various percutaneous

endovascular techniques [6, 7]. Also, injection of embolic material in the fistula followed by endovascular stent graft has been described [8].

Despite the advancements in treatment of aortoduodenal fistulas and state-of-the-art intensive care, the prognosis remains poor, with an overall 30-days survival of 30–44% [4, 5].

Due to the meticulous coverage of the aortic prosthesis after implantation by closing the aneurysm sac and closing the retroperitoneum, an aortoduodenal fistula usually presents as a late complication after transperitoneal open aneurysm repair with an incidence of 0.4–2.4%, mostly 3–5 years after surgery [4]. Mechanisms of secondary fistulas include direct mechanical erosion of the suture line into the bowel, proximal suture line disruption with pseudoaneurysm formation and fistulization, transient bacteremia and graft infection from perioperative contamination [4].

The patient in this case was diagnosed with an aortoduodenal fistula only six weeks after surgery. An aortoduodenal fistula seldom occurs this early after transperitoneal open aneurysm repair. In Table 1, the postoperative time of presentation of an aortoduodenal

or enteric fistula is given [9–25]. Chang et al. described a patient in 2002 who presented 20 days after emergency surgery because of a ruptured abdominal aneurysm with an aortoduodenal fistula [16]. The perioperative cultures were positive for *Klebsiella pneumoniae*. Tromp et al. also described a patient presenting with an early aortoduodenal fistula six weeks after an endovascular aneurysm repair was performed [23].

In this patient, the early occurrence of an aortoduodenal fistula may be explained by the perioperative duodenal serosal injury or by the inflammatory character of the aneurysm. On the preoperative computed tomography scan, it was not expected that an inflammatory aortic aneurysm was present in the patient. Best on based available evidence, if an inflammatory aneurysm had been expected, an open retroperitoneal approach may have been considered [26, 27]. A search of literature according to the Patient Intervention Comparison Outcome (PICO) strategy and critical appraisal method in line with Guyatt [28] and the Cochrane Handbook for systematic reviews [29], showed that repair of an inflammatory aneurysm may have a higher risk of developing an aortoduodenal fistula [16, 30].

Table 1: Reports of secondary aortoduodenal fistulas since 1998 and the time of presentation after abdominal aortic aneurysm repair

Author	Year of Publication	Number of Patients	Time of Fistula after Initial Procedure	Type of Procedure
Yabu [9]	1998	1	10 years	Conventional repair
Constans [10]	1999	7	4-7 years	Conventional repair
Hauseggers [11]	1999	1	20 months	Endovascular repair
Makar [12]	2000	1	4 months	Endovascular repair
Lau, [13]	2001	1	5 years	Conventional repair
Ohki, [14]	2001	1	9 months	Endovascular repair
Parry, [15]	2001	1	12 months	Endovascular repair
Chang, [16]	2002	1	20 days	Conventional repair
Kar, [17]	2002	1	22 months	Endovascular repair
Berges, [18]	2003	1	2 years	Endovascular repair
Elkouri, [19]	2003	1	17 months	Endovascular repair
Probst, [20]	2006	1	8 months	Conventional repair
Geraci [21]	2008	1	5 years	Conventional repair
Tanaka [22]	2009	1	13 years	Conventional repair
Tromp [23]	2009	1	6 weeks	Endovascular repair
Billi [24]	2012	1	3 years	Conventional repair
Perencevich [25]	2013	1	3 years	Conventional repair

CONCLUSION

In summary, an aortoduodenal fistula is a devastating condition, associated with high morbidity and mortality, generally, presenting years after the aneurysm repair. The presented case of an aortoduodenal fistula occurred only six weeks after conventional aneurysm repair. The early development of this aortoduodenal fistula may be explained by the inflammatory character of the abdominal aortic aneurysm.

Author Contributions

Denise MD Özdemir-van Brunschot – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Giel G Koning – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

J Adam van der Vliet – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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