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CASE REPORT

Secondary Aorto-enteric Fistula and Type II Endoleak Five Years after Endovascular Abdominal Aortic Aneurysm Repair

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Introduction: Secondary aorto-enteric fistula (AEF) after endovascular abdominal aortic aneurysm repair (EVAR) is a rare but potentially fatal disease. The aetiology and mechanisms are unclear. This study presents a patient who developed secondary AEF and type II endoleak five years after EVAR.
Case: A 73 year old man underwent successful EVAR with a bifurcated aortic stent graft for a 5.5 cm infrarenal abdominal aortic aneurysm. The aneurysm sac showed no change in size for three years, then shrank 20 mm to 3.5 cm by five years. After five years and eight months, the patient presented with fever and back pain. Enhanced CT demonstrated enlargement of the aneurysm sac, type II endoleak from the third and fourth right lumbar arteries, and air around the stent graft. An emergency operation was performed. The infected stent graft was removed by pushing up the stent graft to release the hooks from the wall of the aorta. A small fistula resembling a fish mouth measuring 1×1 cm was observed in the third part of the duodenum. The fistula was closed by direct suture, and in situ reconstruction was performed with an 18×9 mm standard polyethylene terephthalate graft. Culture of the explanted stent graft grew enterobacter. Intravenous antibiotic therapy was continued for six weeks and was stopped after confirming no recurrence of infection with computed tomography and laboratory testing. Two years later, there has been no recurrence of infection.
Conclusion: Long term surveillance is critical because AEF can occur even after initially successful EVAR.

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INTRODUCTION

Since 1998, secondary aorto-enteric fistula (AEF) has been reported as a rare but potentially fatal disease occurring after endovascular abdominal aortic aneurysm repair (EVAR). The aetiology and mechanisms of secondary AEF remain unclear, and several causes have been reported.[1–24] This study presents a patient who developed secondary AEF with type II endoleak five years after EVAR.

CASE

A 73 year old man underwent successful EVAR with a bifurcated aortic stent graft (Excluder; W. L. Gore and Associates Inc., Newark, DE) for a 5.5 cm infrarenal abdominal aortic aneurysm (AAA) following the manufacturer's instruction for use. He had a previous medical history of hypertension. The post-operative course was uneventful and enhanced CT showed no migration or endoleak one week after EVAR. Follow up with enhanced CT revealed no change in the aneurysm sac with a diameter of 5.5 cm for the first three years, no endoleak and no stent graft migration. After three years, the aneurysm sac started shrinking rapidly to 4.0 cm at four years and 3.5 cm at five years.

After five years and eight months, the patient suffered from fever and back pain with C reactive protein elevated to 20.7 mg/dL and the white blood cell count 19.2×10^9/L. Blood culture revealed no organisms. Enhanced CT demonstrated enlargement of the aneurysm sac to 4.5 cm and type II endoleak from the third and fourth right lumbar arteries (Fig. 1A and B). Air was seen around the stent graft inside the aneurysm sac, and the margin between the duodenum and AAA was ill defined.

Diagnosis of stent graft infection was made, and intravenous antibiotic therapy with meropenem and vancomycin was started. However, as CT showed ventral protrusion of the aneurysm sac two days later (Fig. 2), an emergency operation was indicated.

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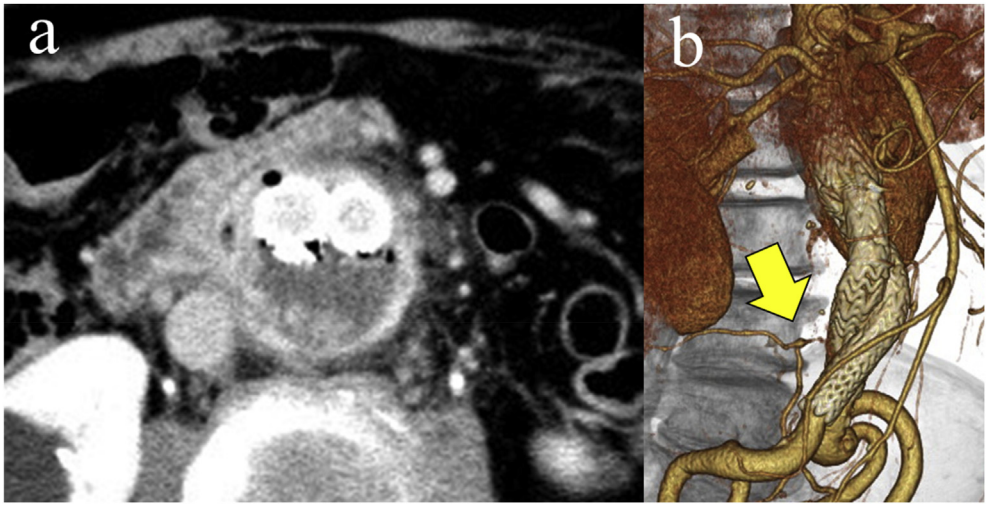

Figure 1. Delayed enhanced CT on re-admission (A) demonstrated type II endoleak from third lumber artery (A). 3D reconstruction (B) (yellow arrows show third lumber artery).

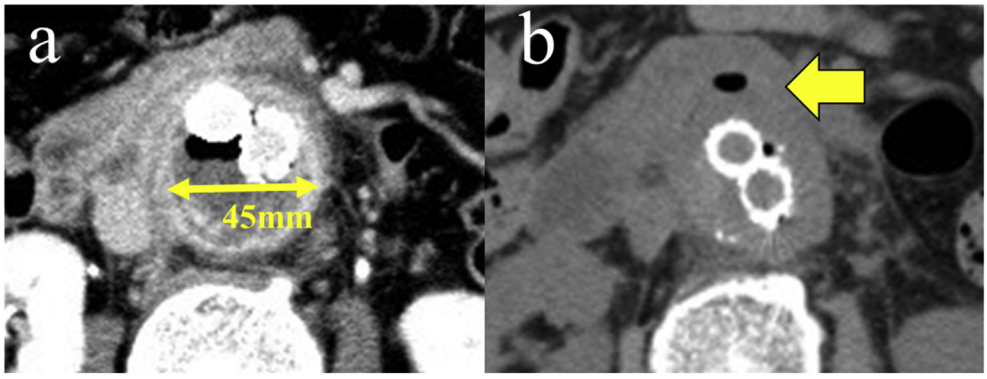

Figure 2. Delayed enhanced CT at five years and eight months on re-admission (A, on re-admission). Two days after readmission (B), ventral protrusion of the aneurysm sac was confirmed.

Through a median laparotomy, the AAA was exposed, and grey malodorous pus was encountered when the aneurysm sac was dissected from the adhesive duodenum. The infrarenal abdominal aorta was clamped above the stent graft, and the common iliac arteries were clamped below the limbs of the stent graft. The stent graft was then wholly removed after opening the aortic sac, and the proximal hooks were removed from the wall of the aorta by pushing the stent graft upwards. Backflow from the third and fourth right lumbar arteries was confirmed. The aneurysm sac was radically debrided together with the surrounding tissue. In the third part of the duodenum there was a small fistula resembling the mouth of a fish measuring 1×1 cm (Fig. 3). The fistula was located away from the anchors at the proximal edge of the Excluder.

The duodenal fistula was closed by direct suture and in situ reconstruction was performed with an 18×9 mm standard polyethylene terephthalate graft (J-graft, Japan Lifeline Co. Ltd., Tokyo, Japan). After vigorous lavage the laparotomy was closed temporarily. The next day, omentopexy was performed, which consisted of wrapping the synthetic prosthesis and covering the suture line on the duodenum with the omental pedicle. The culture of the explanted stent graft revealed only gram negative enterobacter. A fungal test was negative. Intravenous antibiotic therapy with sulbactam/ampicillin and levofloxacin was continued for six weeks. Antibiotic therapy was stopped after confirming no recurrence of infection on computed tomography and laboratory testing. Two years later there has been no recurrence of infection. Informed consent for research was obtained at the outpatient clinic.

DISCUSSION

Secondary AEF after EVAR was reported first by Norgren in 1998.[1] Subsequently, a recent multicentre study from Italy, the MAEFISTO study, reported the incidence of secondary AEF after EVAR as 0.8% for the total cohort, 0.46% (15/3262) for atherosclerotic aneurysmal disease group, and 3.9% (7/179) for the post-surgical pseudoaneurysm group.[25]

Review of the literature yielded 24 previous studies, conducted between 1998 and 2018, which dealt with 32 patients with secondary AEF after EVAR.[1–24] Characteristics and peri-operative data obtained from case reports are

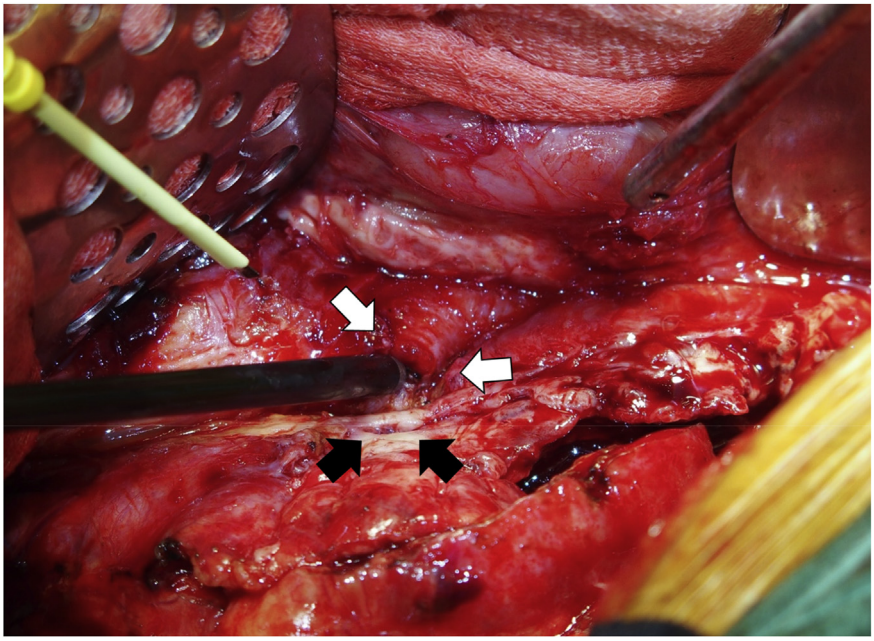

Figure 3. Intra-operative findings revealed a fistula in the third part of the duodenum (white arrows) and a defect in the aneurysm sac (black arrows).

shown in Table 1. The mean age of the 32 patients was 68.7 ± 8.3 y, and 30 (93.8%) were male. The aetiology of AAA was inflammatory for two patients, Behçet's disease for one, and non-specified for 29 patients. The mean interval from EVAR to the diagnosis of secondary AEF was 20.4 ± 17.5 months.

In the present case, the detected micro-organism was the gram negative enterobacter group, and a fungal test was negative. Yeast like fungus was detected from the tissue culture in only one case from the case report review (multiple micro-organisms in seven, gram positive bacterium only in seven, gram negative bacterium in three, only *Bacteroides fragilis* and negative in one, data were not available for 13). However, Batt et al. reported that *Candida* was the most commonly isolated organism from infected grafts in 37 cases of secondary AEF and the frequency was 14 cases (42%).[26] Fungal infections should always be kept in mind as a cause of secondary AEF because fungal infection can lead to poor prognosis because of other organ infection or compromised patient condition.[27]

Several hypotheses have been reported for the mechanisms of fistula formation after EVAR. First, erosion or perforation in the aortic wall may contribute to fistula formation. This mechanical mechanism is caused by the stent graft body with or without migration or kinking, or by the hook, a guidewire, and/or coil.[1,2,20,23] Second, peri-aortic inflammation which includes pre-EVAR infected or inflammatory AAA, local infection, stent graft, and/or intervention for endoleak can lead to oedematous or destructive changes in the aortic and enteric wall.[4,6,9,10,14,17] Third, compression caused by the pre-existing aneurysm sac, endotension, and re-expansion of the aneurysm sac may trigger the erosion of the enteric wall.[2,7,11,16] In addition, perforation of a primary duodenal ulcer can cause secondary AEF, and a combination of all or any of these causes should be taken into consideration.

In the case reported here, the aneurysm sac showed no change in size for three years, had shrunk by five years post-operatively, and then showed secondary AEF, re-expansion of the aneurysm sac, and type II endoleak. The consequence of changes in the aneurysm sac and AEF formation led to speculation that erosion of the enteric wall arose from compression by the aneurysm sac before EVAR and/or the re-expanding aneurysm sac. The influence of type II endoleak in development of sac expansion and secondary AEF cannot be excluded. Several reports have stated that delayed type II endoleak identified one year or more after initial EVAR is associated more with aneurysm sac expansion than early type II endoleak identified within one year.[28,29]

As for the surgical treatment of the secondary AEF, resection of the AAA and stent graft was indicated in 23 cases (72%) followed by an extra-anatomical bypass in 14 and by in situ replacement in nine (polyethylene terephthalate graft in three patients, rifampicin soaked polyethylene terephthalate graft in two patients, arterial allograft, polytetrafluoroethylene graft, silver impregnated graft and rolled pericardial graft in one patient). In these cases, surgical procedures for the enteric fistula entailed suturing in 11 cases, segmental resection of jejunum or ileum in four, gastrostomy and jejunostomy tube in two, omentopexy in one, and unknown in five.

Standard management of secondary AEF after EVAR is explantation of the graft, debridement of the infected surrounding tissue, revascularisation, and surgical intervention for the bowel defect, whenever possible with autologous tissue coverage using omentum. However, the option of revascularisation for secondary AEF is controversial.

Table 1. Characteristics and peri-operative data obtained from case reports.

Patient's munber	Year	Author	Gender	Age, y	Used stent graft	Time after EVAR, mo
1	1998	Norgren	M	70	Stentor	18
2	1999	Housegger	M	52	Vanguard	20
3	2000	Janne d'Othée	M	62	Stentor	22
4	2000	Makar	M	70	Zenith	4
5	2001	Ohki	M	Unknown	Unknown	9
6	2001	Ohki	M	Unknown	Unknown	30
7	2001	Parry	M	61	AneuRx	6
8	2002	Kar	M	76	AneuRx	23
9	2003	Alankar	M	76	AneuRx	4
10	2003	Elkouri	F	75	Talent	17
11	2003	Bertges	M	79	Ancure	53
12	2003	Abou-Zamzam	M	67	Ancure	11
13	2004	French	F	68	Zenith	18
14	2006	Ghosh	M	52	AneuRx	9
15	2006	Ueno	M	69	Custom made	19
16	2007	Ruby	M	76	Ancure	58
17	2008	Saratzis	M	69	Anaconda	6
18	2008	Saratzis	M	75	EndoFit	11
19	2008	Saratzis	M	70	Powerlink	4
20	2008	Saratzis	M	68	EndoFit	1
21	2008	Saratzis	M	60	EndoFit	6
22	2009	Cheu	M	67	Zenith	14
23	2009	Lane	M	69	Excluder	6
24	2009	Riera del Moral	M	62	Talent + FF	46
25	2009	Riera del Moral	M	61	AneuRx	12
26	2009	Riera del Moral	M	73	Talent	2
27	2012	Benjamin	M	66	Excluder	36
28	2012	Farres	M	76	AneuRx	46
29	2014	Zaki	M	75	Endurant II	Unknown
30	2018	Arworn	M	42	Endurant II	13
31	2018	Walter	M	75	Endurant II	48
32	2018	Present	M	73	Excluder	60

Aetiology of AEF	Symptoms	Treatments	Results
Graft rupture	Abdominal pain, bowel haemorrhage	*in situ* replacement	Alive at six months
Graft migration and kinking	Abdominal pain, bowel haemorrhage	*in situ* replacement	Alive at six months
Graft migration and kinking	Fever, bowel haemorrhage	Extra-anatomical bypass	Alive at 40 months
Crohn's disease	Fever, abdominal pain, melaena	Antibiotics therapy alone	Died
Unknown	Bowel haemorrhage	Unknown	Died
Infection	Fever	Antibiotics therapy alone	Died
Inflammatory	Fever, back pain, vomiting	*in situ* replacement	Alive at seven months
Endotension	Fever	*in situ* replacement	Alive at one year
Type Ia endoleak	Abdominal pain, melaena	*in situ* replacement	Alive at six months
Coil embolisation	Fever, vomiting, anorexia	Extra-anatomic bypass	Died
Coil embolisation	Fever, vomiting	Extra-anatomic bypass	Alive at one month
Endotension	Abdominal pain, vomiting	Extra-anatomical bypass	Alive at four months
Infection	Haematemesis, melaena	Extra-anatomical bypass	Died
Infection	Fever	None	Died
Unknown	Abdominal pain	Extra-anatomical bypass	Alive at 15 months
Endotension	Abdominal pain, nausea	*in situ* replacement	Alive at 13 months
Unknown	Abdominal pain, haematemesis	Unknown	Died
Type I endoleak	Abdominal pain, haematemesis	None	Died
Unknown	Abdominal pain, haematemesis	Unknown	Died
Infection	Abdominal pain, haematemesis	Extra-anatomical bypass	Alive at three years
Infection	Abdominal pain, haematemesis	Extra-anatomical bypass	Alive at one year
Endotension	Fever, lumber pain	*in situ* replacement	Alive at two months
Unknown	Fever, diarrhoea	Extra-anatomical bypass	Alive at two weeks
Unknown	Melaena	Extra-anatomical bypass	Died
Unknown	Fever, back pain, haematemesis	Repair by pericardium patch	Died
Type Ia endoleak	Melaena	Antibiotics therapy alone	Died

Continued

Table 1-*continued*

Aetiology of AEF	Symptoms	Treatments	Results
Infection	Melaena	Extra-anatomical bypass	Alive at three months
Guidewire perforation	No description	Extra-anatomical bypass	Alive at one year
Infection	Abdominal pain, haematemesis	Extra-anatomical bypass	Died
Graft migration and kinking	Abdominal pain	Extra-anatomical bypass	Alive at nine months
Infection	Fever, abdominal pain	*in situ* replacement	Alive at five mo
Unknown	Fever, back pain	*in situ* replacement	Alive at one year

EVAR = endovascular abdominal aortic aneurysm repair, AEF = aorto-enteric fistula.

Recently, excellent outcomes have been reported after in situ replacement in a multicentre study.[30] The extra-anatomical bypass is usually chosen when the graft or surrounding tissue is highly contaminated with a purulent fluid collection or gross retroperitoneal infection. This technique must be performed carefully to prevent aortic stump disruption and thrombosis, and graft occlusion.[25] Surgical intervention for enteric fistula is an integral part in both in situ reconstruction and extra-anatomical bypass. Simple closure of the enteric fistula may be performed when the defect is small. If the defect is large, segmental bowel resection should be performed to prevent leakage resulting in recurrent infection.

The possible materials resistant to infection are arterial allograft and deep femoral vein graft.[30] Nevertheless, antibiotic soaked graft or silver coated graft may be used for in situ reconstruction.

Omentopexy after resection of the AAA and stent graft was performed in 11 cases (48%). In the present case, omentopexy was performed the day after in situ replacement to prevent recurrent infection. The advantage of staged omentopexy includes certainty about haemostasis, meticulous lavage, and also surgical instruments contaminated by bacteria are replaced with sterilised instruments. Hospital mortality after secondary AEF was 17.4% (4/23), which is similar to that reported by a multicentre study.[25]

CONCLUSION

This report was of the development of secondary AEF and type II endoleak over five years after successful EVAR. Long term surveillance is critical as AEF can occur even after initially successful EVAR.

CONFLICT OF INTEREST

None.

FUNDING

None.

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