CASE REPORT

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

Yojiro Koda a, Hirohisa Murakami b, Masato Yoshida b, Hitoshi Matsuda c,*, Nobuhiko Mukohara b

a Division of Cardiovascular Surgery, Department of Surgery, Kobe University Hospital, Hyogo, Japan
b Department of Cardiovascular Surgery, Hyogo Brain and Heart Centre, Himeji, Japan
c Department of Cardiovascular Surgery, National Cerebral and Cardiovascular Centre, Suita, Japan

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. © 2019 The Authors. Published by Elsevier Ltd on behalf of European Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Keywords: Endovascular abdominal aortic aneurysm repair, Secondary aorto-enteric fistula, Type II endoleak

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 19.2×10⁹/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.
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
shown in Table 1. The mean age of the 32 patients was 68.7 ± 8.3 years, 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%). 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.

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. 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. 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. 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.

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 number | Year | Author | Gender | Age, y | Used stent graft | Time after EVAR, mo |
|------------------|------|--------|--------|--------|------------------|-------------------|
| 1                | 1998 | Norgren M | M | 70 | Stentor | 18 |
| 2                | 1999 | Housegger M | M | 52 | Vanguard | 20 |
| 3                | 2000 | Janne d'Othée M | M | 62 | Stentor | 22 |
| 4                | 2000 | Makar M | M | 70 | Zenith | 4 |
| 5                | 2001 | Ohki M | M | Unknown | Unknown | 9 |
| 6                | 2001 | Ohki M | M | Unknown | Unknown | 30 |
| 7                | 2001 | Parry M | M | 61 | AneuRx | 6 |
| 8                | 2002 | Kar M | M | 76 | AneuRx | 23 |
| 9                | 2003 | Alankar M | M | 76 | AneuRx | 4 |
| 10               | 2003 | Elkouri F | F | 75 | Talent | 17 |
| 11               | 2003 | Bertges M | M | 79 | Ancure | 53 |
| 12               | 2003 | Abou-Zamzam M | M | 67 | Ancure | 11 |
| 13               | 2004 | French F | F | 68 | Zenith | 18 |
| 14               | 2006 | Ghosh M | M | 52 | AneuRx | 9 |
| 15               | 2006 | Ueno M | M | 69 | Custom made | 19 |
| 16               | 2007 | Ruby M | M | 76 | Ancure | 58 |
| 17               | 2008 | Saratzis M | M | 69 | Anaconda | 6 |
| 18               | 2008 | Saratzis M | M | 75 | EndoFit | 11 |
| 19               | 2008 | Saratzis M | M | 70 | Powerlink | 4 |
| 20               | 2008 | Saratzis M | M | 68 | EndoFit | 1 |
| 21               | 2008 | Saratzis M | M | 60 | EndoFit | 6 |
| 22               | 2009 | Cheu M | M | 67 | Zenith | 14 |
| 23               | 2009 | Lane M | M | 69 | Excluder | 6 |
| 24               | 2009 | Riera del Moral M | M | 62 | Talent + FF | 46 |
| 25               | 2009 | Riera del Moral M | M | 61 | AneuRx | 12 |
| 26               | 2009 | Riera del Moral M | M | 73 | Talent | 2 |
| 27               | 2012 | Benjamin M | M | 66 | Excluder | 36 |
| 28               | 2012 | Farres M | M | 76 | AneuRx | 46 |
| 29               | 2014 | Zaki M | M | 75 | Endurant II | Unknown |
| 30               | 2018 | Arworn M | M | 42 | Endurant II | 13 |
| 31               | 2018 | Walter M | M | 75 | Endurant II | 48 |
| 32               | 2018 | Present M | 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     |
| Crohn’s disease                   | Fever, bowel haemorrhage        | Extra-anatomical bypass  | Alive at 40 months      |
| Inflammation                      | Fever, back pain, vomiting      | *in situ* replacement     | Alive at seven months   |
| Endotension                       | Fever                           | *in situ* replacement     | Alive at one year       |
| Type I 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    |
| Endotension                       | Abdominal pain                   | *in situ* replacement     | Alive at one year       |
| Unknown                           | 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 I endoleak                   | Melaena                         | Antibiotics therapy alone | Died                    |

Continued
Recently, excellent outcomes have been reported after in situ replacement in a multicentre study. 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. 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. 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.

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.

REFERENCES
1. Norgren L, Jernby B, Engellau L. Aortoenteric fistula caused by a ruptured stent-graft: a case report. J Endovasc Surg 1998;5: 269–72.
2. Hausegger KA, Tiesenhausen K, Karaic R, Tauss J, Koch G. Aortoenteric fistula: a late complication of intraluminal exclusion of an infrarenal aortic aneurysm. J Vasc Interv Radiol 1999;10:747–50.
3. Janne d’Othée B, Soula P, Otal P, Cahill M, Joffre F, Cérène A, et al. Aortoduodenal fistula after endovascular stent-graft of an abdominal aortic aneurysm. J Vasc Surg 2000;31:190–5.
4. Makr R, Reid J, Pherwani AD, Johnston LC, Hannon RJ, Lee B, et al. Aorto-enteric fistula following endovascular repair of abdominal aortic aneurysm. Eur J Vasc Endovasc Surg 2000;20:588–90.
5. Ohki T, Veith FI, Shaw P, Lipsitz E, Suggs WD, Wain RA. Increasing incidence of mid-term and long-term complications after endovascular graft repair of abdominal aortic aneurysms: a note of caution based on a 9-year experience. Ann Surg 2001;234:323–35.
6. Parry DJ, Waterworth A, Kessel D, Robertson I, Berridge DC, Scott DJ. Endovascular repair of an inflammatory abdominal aortic aneurysm complicated by aortoenteric fistula with an unusual presentation. J Vasc Surg 2001;33:874–9.
7. Kar B, Dougherty K, Reul GJ, Krajcer Z. Aortic stent-graft infection due to a presumed aortoenteric fistula. J Endovasc Ther 2002;9:901–6.
8. Alankar S, Barth MH, Shin DD, Hong JR, Rosenberg WR. Aortoenteric fistula and associated rupture of abdominal aortic aneurysm after endoluminal stentgraft repair. J Vasc Surg 2003;37:465–8.
9. Elkouri S, Blair JF, Therasse E, Oliva VL, Bruneau L, Soulez G. Aortoenteric fistula occurring after type II endoleak treatment with coil embolization of the aortic sac. J Vasc Surg 2003;37:461–4.
10. Bertges DJ, Villella ER, Makaroun MS. Aortoenteric fistula due to endoleak coil embolization after endovascular AAA repair. J Endovasc Ther 2003;10:130–5.
11. Abou-Zamzam AM, Bianchi C, Mazraany W, Teruya TH, Hopewell J, Vannix RS. Aortoenteric fistula development following endovascular abdominal aortic aneurysm repair: a case report. Ann Vasc Surg 2003;17:119–22.
12. French JR, Simring DV, Merrett N, Thursby P. Aorto-enteric fistula following endoluminal abdominal aortic aneurysm repair. ANZ J Surg 2004;74:397–9.
13. Ghosh J, Murray D, Khwaja N, Murphy MO, Halka A, Walker MG. Late infection of an endovascular stent graft with septic embolization, colonic perforation, and aortoduodenal fistula. Ann Vasc Surg 2006;20:263–6.
14. Ueno M, Iguro Y, Nagata T, Sakata R. Aortoenteric fistula after endovascular stent grafting for an abdominal aortic aneurysm: report of a case. Surg Today 2006;36:546–8.
15. Ruby BJ, Cogbill TH. Aortoduodenal fistula 5 years after endovascular abdominal aortic aneurysm repair with the Ancure stent graft. J Vasc Surg 2007;45:834–6.
16. Chenu C, Marcheix B, Barcelo C, Rousseau H. Aorto-enteric fistula after endovascular abdominal aortic aneurysm repair: case report and review. Eur J Vasc Endovasc Surg 2009;39:401–6.
17. Saratzis N, Saratzis A, Melas N, Ktenidis K, Kiskinis D. Aorto-duodenal fistulas after endovascular stent-graft repair of
abdominal aortic aneurysms: single-center experience and review of the literature. J Endovasc Ther 2008;15:441–8.

18 del Moral LR, Alonso SF, Kiuri SS, Caballero DF, Heredero AF, Nistal MG, et al. Aortoenteric fistula arising as a complication of endovascular treatment of abdominal aortic aneurysm. Ann Vasc Surg 2009;23:255.

19 Lane JS, Barleben AR, Kubaska SM, Fujitani RM. Aortoenteric fistula arising as a complication of endovascular treatment of abdominal aortic aneurysm. Ann Vasc Surg 2009;23:255.

20 Farres H, Gonzales AH, Garrett HE. Aortoduodenal fistula after endograft repair of abdominal aortic aneurysm secondary to a retained guidewire. J Vasc Surg 2012;56:1413–5.

21 Lind BB, Jacobs CE. Primary aortoduodenal fistula supplied by type II endoleak. Ann Vasc Surg 2012;26:1012.

22 Zaki M, Tawfick W, Alawy M, ElKassaby M, Hynes N, Sultan S. Secondary aortoduodenal fistula following endovascular repair of inflammatory abdominal aortic aneurysm due to Streptococcus anginosus infection: a case report and literature review. Int J Surg Case Rep 2014;5:710–3.

23 Arworn S, Orrapin S, Chakrabandhu B, Reanpang T, Settakorn J, Laohapensang K. Aorto-enteric fistula after endovascular abdominal aortic aneurysm repair for Behcet’s disease patient: a case report. RVES Short Rep 2018;39:54–7.

24 Walter C, Taher F, Rieger H, Assadjan A, Falkensammer J. Endograft infection due to secondary aortoenteric fistula treated with custom-made bovine in situ aortic bifurcation graft. Vasc Endovasc Surg 2018;18:1–5.

25 Kahlberg A, Rinaldi E, Piffaretti G, Speziale F, Trimarchi S, Bonardelli S, et al. MAEFISTO collaborators. Results from the multicenter study on aortoenteric fistulization after stent grafting of the abdominal aorta (MAEFISTO). J Vasc Surg 2016;64:313–20.

26 Batt M, Jean-Baptiste E, O’Connor S, Saint-Lebes B, Feugier P, Patra P, et al. Early and late results of contemporary management of 37 secondary aortoenteric fistulae. Eur J Vasc Endovasc Surg 2011;41:748–57.

27 Končar IB, Dragaš M, Sabljak P, Peško P, Marković M, Davidović L. Aortoesophageal and aortobronchial fistula caused by Candida albicans after thoracic endovascular aortic repair. Vojnosanit Pregl 2016;73:684–7.

28 Zhou W, Blay E, Varu V, Ali S, Jin MQ, Sun L, et al. Outcome and clinical significance of delayed endoleaks after endovascular aneurysm repair. J Vasc Surg 2014;59:915–20.

29 Pineda DM, Calligaro KD, Tyagi S, Troutman DA, Dougherty NH. Late type II endoleaks after endovascular aneurysm repair require intervention more frequently than early type II endoleaks. J Vasc Surg 2018;67:449–52.

30 Smeds MR, Duncan AA, Harlander-Locke MP, Lawrence PF, Lyden S, Fatima J. Vascular Low-Frequency Disease Consortium. Treatment and outcomes of aortic endograft infection. J Vasc Surg 2016;63:332–40.