Turkish Journal of Thoracic and Cardiovascular Surgery 2021;29(3):399-403
http://dx.doi.org/doi: 10.5606/tgkdc.dergisi.2021.21518

Case Report / Olgu Sunumu

Surgical approach to recurrent secondary aortoenteric fistulas: A case report

Tekrarlayan sekonder aortoenterik fistüllerde cerrahi yaklaşım: Olgu sunumu

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ABSTRACT

The increasing number of abdominal aortic grafts due to abdominal aortic aneurysms has caused secondary aortoenteric fistulas to be seen more frequently as a cause of gastrointestinal bleeding. High index of suspicion plays a significant role in the diagnosis in patients having clinical symptoms ranging from fecal occult blood to massive gastrointestinal bleeding, accompanied by hemorrhagic shock. A 65-year-old male patient developed two secondary aortoenteric fistulas consecutively. The first one was aortic graft-jejunal and the second one was aortic graft-duodenal in a short period. Secondary aortoenteric fistula developed after aortobifemoral bypass. The patient underwent graft revision and jejunal repair. He was reoperated three months later due to the newly developed aortic graft-duodenal fistula. The duodenal defect was closed, and an extra-anatomic aortoiliac bypass was performed to avoid graft-related enteric fistula. The patient was discharged uneventfully and was free from any complication at nine months after surgery.

Keywords: Extra-anatomic reconstruction, graft-duodenal fistula, graft-jejunal fistula, recurrent graft enteric fistula, secondary aortoenteric fistula.

ÖZ

Abdominal aort anevrizmasına bağlı artan abdominal aort greft sayısı, sekonder aortoenterik fistüllerin gastrointestinal kanama nedeni olarak daha sık görülmesine neden olmuştur. Gaytada gizli kandan masif gastrointestinal kanamaya kadar değişen ve hemorajik şokun da eşlik ettiği klinik semptomları olan hastalarda yüksek düzeyde şüphe tanıda önemli bir rol oynar. Altmış beş yaşında erkek hastada ardışık olarak iki sekonder aortoenterik fistül gelişti. Bunlardan birincisi kısa sürede aort greft-jejunal ve ikincisi aort grefti-duodenal fistül idi. Hastada aortobifemoral baypas sonrası sekonder aortoenterik fistül gelişti. Hastaya greft revizyonu ve jejunal onarım yapıldı. Yeni gelişen aort grefti-duodenal fistül nedeniyle üç ay sonra tekrar ameliyat edildi. Duodenal defekt kapatıldı ve greftin tekrar enterik fistül geliştirmemesi için ekstra anatomik aortoiliyak baypas yapıldı. Hasta sorunsuz bir şekilde taburcu edildi ve ameliyatın dokuzuncu ayında herhangi bir komplikasyon izlenmedi.

Anahtar sözcükler: Ekstra anatomik rekonstrüksiyon, greft-duodenal fistül, greft-jejunal fistül, tekrarlayan greft enterik fistül, sekonder aortoenterik fistül.

Secondary aortoenteric fistulas (SAEFs) occurs in 0.4 to 2.4% following abdominal aortic reconstruction (AAR) and is treated with high morbidity/mortality.[1] More fistulas occur in the third part of the duodenum located between the abdominal aortic and the superior mesenteric artery.[2] The development of SAEFs should be suspected in patients operated due to AAR and those presenting with complaints such as gastrointestinal bleeding (GIB), a pulsatile mass in the abdomen, and infection.[3] Although endoscopy and computed tomography (CT) angiography are usually helpful in the diagnosis, some fistulas can be only detected intraoperatively in the intestinal segment. Surgical

Received: February 01, 2021 *Accepted*: April 12, 2021 *Published online*: July 26, 2021

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Cite this article as:
Zarbaliyev E, Konukoğlu O, Çağlıkülekçi M, Mansuroğlu D, Baş S, Kilercik H. Surgical approach to recurrent secondary aortoenteric fistulas: A case report. Turk Gogus Kalp Dama 2021;29(3):399-403

treatment is the preferred option, and emergency treatment is needed in selected cases.

In the literature, a case report was reported due to an SAEF treated with endovascular treatment and developed SAEF again four months later.[4] In this article, we present a 65-year-old male patient with GIB complaints and a history of SAEF twice. In this respect, our case is rare due to the development of SAEF twice after open surgery and in a short time.

CASE REPORT

A 65-year-old male patient with intermittent claudication and exertional distal calf pain was diagnosed with aortoiliac obstructive peripheral vascular disease. Aortobifemoral bypass was applied from the infrarenal level with a polyester intervascular (18/9 mm) bifurcated graft (Maquet Getinge Group, Rastatt, Germany) 16 months ago. He was discharged after a week of follow-up. He was admitted to the emergency department 14 months later due to hematochezia. The patient underwent upper gastrointestinal endoscopy and, examined up to the proximal jejunal loop. No bleeding focus was evident. (Figure 1a). On CT angiography, fistula development between the aortic graft and distal jejunal segment was detected with active bleeding (Figure 1b). The patient was taken to emergency surgery, and during exploration, an infected pseudoaneurysm originating from the superior part of the proximal anastomosis was detected. Also, a fistula was observed between the right iliac arm of the graft and the jejunum.

Aortic cross-clamp was placed infrarenally and, then, the graft-jejunal fistula was separated. The jejunal defect was closed using a linear stapler (Figure 2a). The remaining synthetic graft was excised and replaced by a Biointegral Surgical No-React® (Biointegral Surgical Inc., ON, Canada) bovine pericardial xenograft due to the presence of active infection in the retroperitoneal cavity. The graft was wrapped with omentum. Intravenous antibiotics (ampicillin-sulbactam + clindamycin) were started based on graft culture, resulting in Streptococcus anginosus. After 19 days of treatment, the patient was discharged. However, after three months, he was re-admitted to the emergency department with melena. On endoscopic examination, the fistula was seen on the second part of the duodenum (Figure 1c). On CT angiography, a new fistula tract formed between the aortic graft's proximal and the second part of

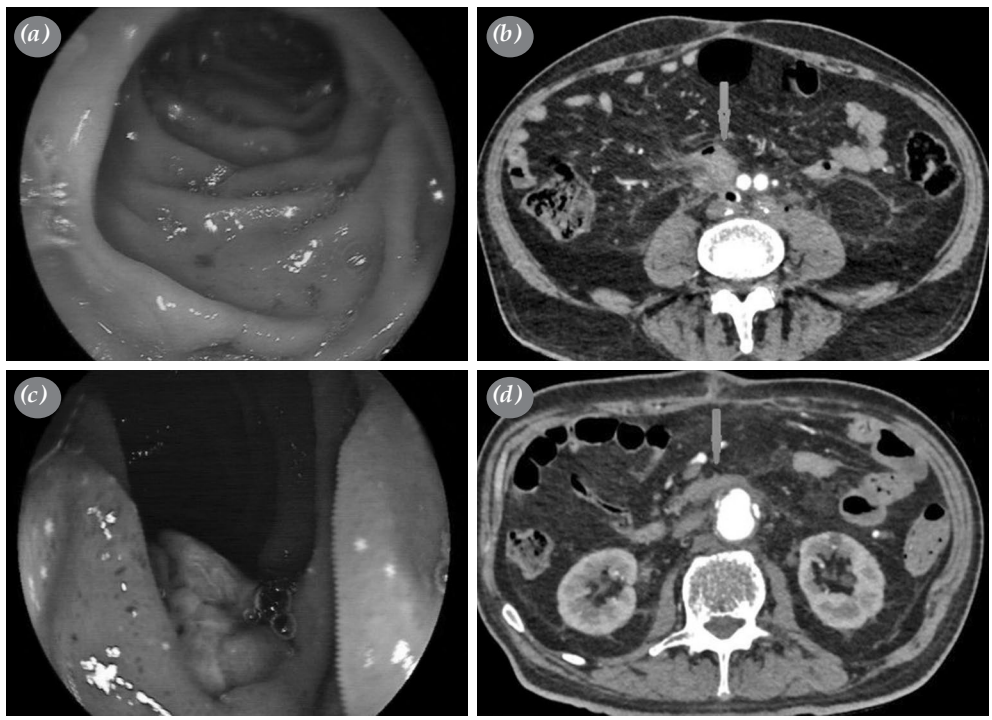

Figure 1. (a) Normal duodenal mucosa. **(b)** Graft-jejunal fistula image on CT angiography. **(c)** View of the fistula mouth in the second part of the duodenum. **(d)** Graft-duodenal fistula image on CT angiography.
CT: Computed tomography.

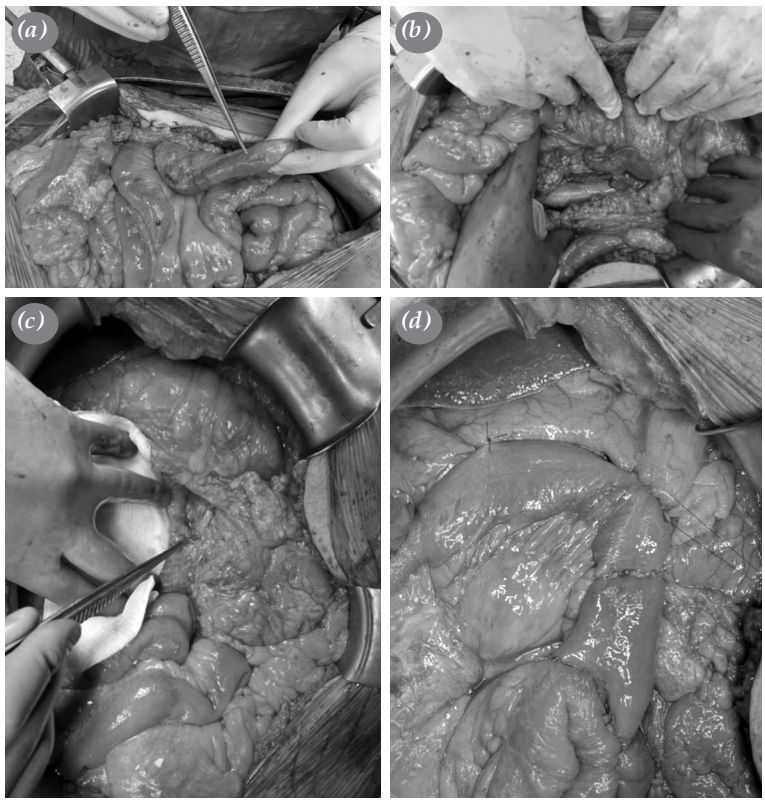

Figure 2. (a) Repaired jejunum segment. **(b)** Repaired duodenal segment. **(c)** Graft-duodenal fistula. **(d)** Gastrojejunostomy for duodenal bypass.

the duodenum and inflammation in the surrounding tissues were observed (Figure 1d). The patient was reoperated three months after his first aortojejunal fistula surgery. Through a left thoracotomy, the aorta was explored to place a cross-clamp, if needed.

By intrabdominal exploration, the fistula from the graft to the second part of the duodenum and infection around surrounding tissues were seen. The duodenal defect was closed with a linear stapler (Figure 2b,c). The Roux-en Y gastrojejunostomy was performed for bypass of the repaired duodenal region (Figure 2d). The aortobifemoral graft was removed and left stumped in the infrarenal area (Figure 3a, b). The stump was stabilized by 4/0 propylene sutures. A side clamp was placed on the thoracic aorta and, proximal anastomosis was made. Extra-anatomically placed graft (12-mm Polyester Maquet Getinge Group, Rastatt, Germany) was anastomosed on the left femoral artery. The graft-to-cross-femoral bypass (10 mm polyester Maquet Getinge Group, Rastatt, Germany) was made to the right femoral artery (Figure 3c). The culture was taken from the infected tissue, *Klebsiella Pneumoniae* was detected, and intravenous ciprofloxacin + linezolid therapy was started. On Day 6, oral food intake was started, and he tolerated it well. There were no problems in the control CT angiography scan. The patient was discharged on Day 14 with oral antibiotic therapy. There was no problem during the follow-up visit at one year. A written informed consent was obtained from the patient.

DISCUSSION

The number of reported case reports of SAEF over the last two decades has been increasing. The fact that many reported cases resulted in mortality show the severity of SAEF.[5] Although patients present with different clinical complaints, upper or upper GIB is the most common cause of admittance. Patients may also present with occult bleeding in routine tests or massive clinical bleeding.[2,6]

The course for the development of SAEF is reported differently in many studies and it usually occurs after a year (range, 1 to 20 years).[2,3] The first SAEF, in our case, occurred 16 months after the initial surgery, consistent with the literature. However, the second SAEF developed within three months. The earliest reported SAEF incidence to date is reported as four months by Colombi et al.[4]

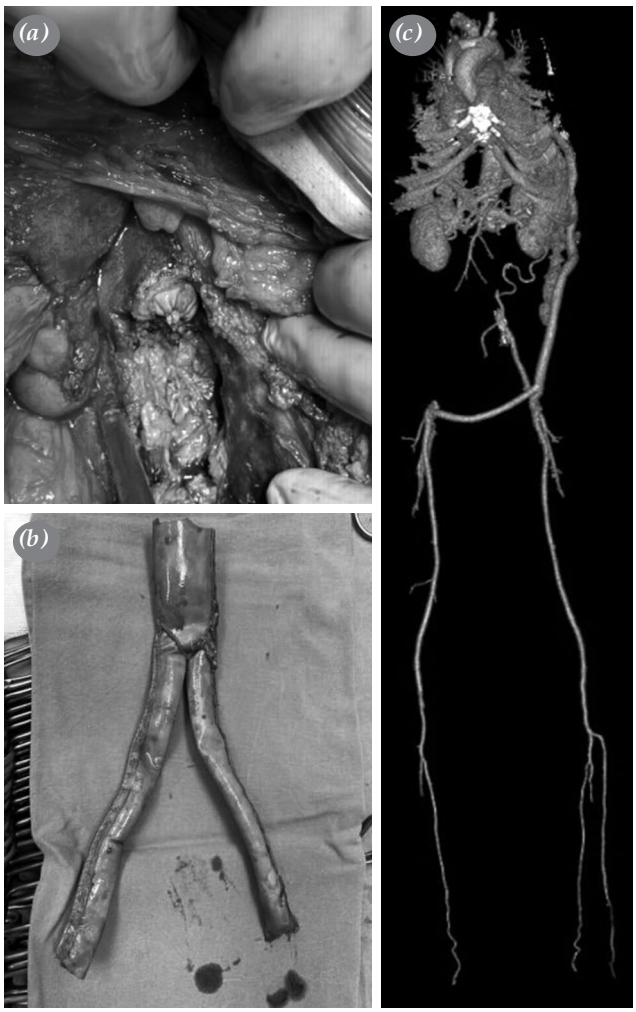

Figure 3. (a) Aortic graft stump. **(b)** Extracted xenograft. **(c)** Extra-anatomic bypass for the abdominal aorta.

Furthermore, SAEF to duodenum after aortic surgery is more common (60 to 88%), but may develop between different regions of the jejunum, ileum, appendix, and large intestine. The more exposure of the duodenum and the jejunum is related to the fact that it is in the proximal aortic anastomosis region, closer to the suture line.[2] Therefore, the creation of barriers consisting of tissues between the graft and intestinal structures following graft implantation has been reported and recommended.[5] Although the third and fourth part of the duodenum was more frequently affected by the placement between the superior mesenteric artery and aorta, the second part of the duodenum was involved in our case. To the best of our knowledge, this is the first case in the literature on this aspect. The development rate of jejunal SAEFs has been reported as ranging from 4 to 24% in the literature[2,5] and, in our case, SAEF was formed in the distal jejunum. However, in the first SAEF, the jejunum was fistulated not with proximal anastomosis, but with pseudoaneurysms.

The mortality of sepsis resulting from graft infection is high, and only antibiotic treatment is insufficient.[7] Mostly isolated specimens are *Enterobacter*, *Escherichia coli*, *Klebsiella*, *Pseudomonas*, *Streptococcus*, and methicillin-resistant *Staphylococcus aureus*.[2,3] In the initial operation, *Streptococcus anginosus* and, then, *Klebsiella* were reproduced in our case and appropriate antibiotic therapy was given.

Endoscopy and CT angiography are the first-line diagnostic methods, as GIB is the most common clinical symptom in these patients.[2,3] The appearance of graft, the presence of active bleeding or clots, and pulsatile mass image are definitive diagnostic endoscopic findings for SAEF.[2,8] In addition, CT angiography helps to identify infection around the aortic graft, the intestinal segment developing fistula, and extravasation of the contrast material. It has been shown that CT angiography has over 90% sensitivity and specificity in these patients.[2,9] In our case, the upper GIS endoscopy was performed, and no bleeding focus was seen. During the first endoscopy, the fistula was at the level of the distal jejunum. During the second admission, a necrotic and emerging fistula was detected in the second part of the duodenum.

For the treatment of SAEFs, it is preferable to perform open surgery and place endovascular stents.[5,10] Stenting is preferred for shorter processing time and avoiding ischemic problems that may occur during aortic clamping. In a multicenter study of Kakkos et al.,[10] the morbidity rate (25%) was lower in the endovascular method in SAEF treatment. In another study, Antoniou et al.[11] reported that 44% of the patients using this method developed re-infection. Of note, the use of open surgery technique has superior aspects, such as removing infected grafts and repairing the intestinal defect. However, the reason for the preference of a biological graft in the first correction surgery in our case was resistance to infection, existing literature data, and our past clinical experience.[12] On the other hand, this case highlights that biological grafts may also cause aortoenteric fistulas in the infected area, revealing the importance of choosing an extra-anatomic bypass directly in patients with SEAFs.

Different extra-anatomic bypass techniques with thoracic aortic and axillary artery have been

reported.[2,5,8] Complications after thoracic artery bypass are less seen (thrombosis due to long graft, motion hoarseness). Therefore, it is preferred more frequently.[5] In our case, we preferred extra-anatomic bypass from thoracic levels. The reason is to use larger grafts, shorten the distance and, thus, increase the graft's flow quality. Batt et al.[3] examined patients who underwent surgery due to SAEF and found no significant difference in morbidity and mortality between *in situ* and extra-anatomic bypasses. Dacron, silver-absorbed graft, and allografts can be used for re-bypass procedures.[3,5] In the first SAEF surgery, we used an infection-resistant *in situ* xenograft (porcine pericardial). The infection did not resolve, and we preferred extra-anatomic supra-diaphragmatic thoracobifemoral bypass in the second surgery. Endovascular interventions are not curative treatment in these patients, due to the high risk of recurrence. However, it can save time in patients with critical conditions.[5,10]

In conclusion, secondary aortoenteric fistulas have high morbidity and mortality rates. Many issues in the treatment of these fistulas are still unclear. Recurrent fistulas are sporadic conditions which are challenging to manage. In these patients, recurrence may develop, leading to gastrointestinal bleeding, even within three months after the revision surgery. In recurrent cases, surgery should not be avoided, and extra-anatomic bypass from the thoracic aorta should be considered.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Moláček J, Třeška V, Čertík B, Baxa J, Houdek K, Opatrný V. An aortoenteric and a graft-enteric fistulae – A severe acute abdominal event. Cor et Vasa 2015;57:e108-e114.
2. Malik MU, Ucbilek E, Sherwal AS. Critical gastrointestinal bleed due to secondary aortoenteric fistula. J Community Hosp Intern Med Perspect 2015;5:29677.
3. 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.
4. Colombi D, Bodini FC, Sverzellati N, Morelli N, Capelli P, Michieletti E. A case of recurrent secondary aortoenteric fistula 4 months after surgery treated by endovascular coiling of the aortic stump and bilateral chimney stent grafts to renal arteries. Ann Vasc Surg 2019;59:310.e1-310.e5.
5. Schoell T, Manceau G, Chiche L, Gaudric J, Gibert H, Tresallet C, et al. Surgery for secondary aorto-enteric fistula or erosion (SAEFE) complicating aortic graft replacement: A retrospective analysis of 32 patients with particular focus on digestive management. World J Surg 2015;39:283-91.
6. Akın M, Gökbayır H, Tezcaner T, Kılıç K, Özdemir E, Ferahköşe Z, et al. Successful treatment of secondary aortoenteric fistula with a Valdoni-Strong's procedure. Turk Gogus Kalp Dama 2011;19:267-9.
7. Herdrich BJ, Fairman RM. How to manage infected aortic endografts. J Cardiovasc Surg (Torino) 2013;54:595-604.
8. Moulton S, Adams M, Johansen K. Aortoenteric fistula. A 7 year urban experience. Am J Surg 1986;151:607-11.
9. Armstrong PA, Back MR, Wilson JS, Shames ML, Johnson BL, Bandyk DF. Improved outcomes in the recent management of secondary aortoenteric fistula. J Vasc Surg 2005;42:660-6.
10. Kakkos SK, Antoniadis PN, Klonaris CN, Papazoglou KO, Giannoukas AD, Matsagkas MI, et al. Open or endovascular repair of aortoenteric fistulas? A multicentre comparative study. Eur J Vasc Endovasc Surg 2011;41:625-34.
11. Antoniou GA, Koutsias S, Antoniou SA, Georgiakakis A, Lazarides MK, Giannoukas AD. Outcome after endovascular stent graft repair of aortoenteric fistula: A systematic review. J Vasc Surg 2009;49:782-9.
12. Terlecki P, Zubilewicz T, Wojtak A, Pleban E, Przywara S, Iłżecki M, et al. Replacement of infected aortoiliac vascular grafts with bifurcated BioIntegral Surgical No-React® bovine pericardial xenografts. Xenotransplantation 2019;26:e12496.