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CASE REPORT



## Secondary aortoenteric fistula successfully treated with staged endovascular repair and duodenal resection without graft removal

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### ABSTRACT

Secondary aorto-enteric fistulae (SAEF) are rare life-threatening complications that occur after abdominal aortic graft implant to treat aortic aneurysm or occlusive disease. Conventional treatments such as extra-anatomic bypass grafting with aortic ligation and subsequent graft removal with bowel repair are associated with a 25% to 90% operative mortality rate. In the emergency setting, patients unsuitable for major arterial surgery may benefit from a staged, less invasive approach. We present a case of SAEF treated with endoluminal deployment of a stent graft followed by duodenojejunal resection and anastomosis without further aortic reconstruction and graft removal.

### ARTICLE HISTORY

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### KEYWORDS

Duodenal resection;  
endovascular repair;  
endovascular stent graft;  
high-risk patients;  
secondary aortoenteric fistula

### Introduction

Secondary aorto-enteric fistulae (SAEF) are rare life-threatening complications that occur after abdominal aortic graft implant to treat aortic aneurysm or occlusive disease. The first case of secondary aorto-enteric fistula (SAEF) was reported in 1953 by Brock, who described direct communication between the proximal anastomosis of an aortic homograft and the duodenum [1]. The incidence of SAEF ranges from 0.3%–2.3% after aortic prosthetic reconstruction [2,3].

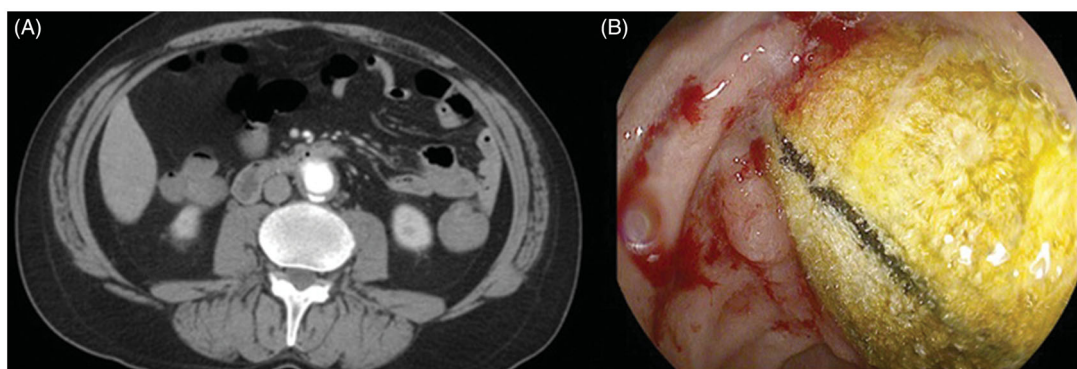
The main objectives of SAEF treatment are maintenance of hemodynamic stability, surgical repair of the underlying defects, infection control via empiric intravenous antibiotics, and perfusion of the lower limbs. The optimum management of SAEF is currently not well defined, and there is a wide range of treatment modalities. These include a variety of open or endovascular procedures, including staged or concomitant extra-anatomical bypass grafting with aortic graft removal and duodenal repair, *in situ* graft replacement, endograft repair with aorto-enteric fistula (AEF) sealing, patching with/without graft removal, and transabdominal drainage [4,5].

When an endovascular stent graft is deployed over an AEF in an acute setting, a second open procedure

is often required to control sepsis and/or treat the enteric defect; additionally, long-lasting antimicrobial therapy protocol is initiated [6,7]. We successfully treated a patient with non-curative surgery for SAEF using a staged endovascular repair and duodenal resection without further aortic reconstruction and graft removal. Here we report the perioperative management and short-term and mid-term results.

### Case presentation

A 61-year-old man with hypertension, chronic obstructive pulmonary disease (COPD), myocardial ischemia, and an aorto-bifemoral bypass graft placed eight years previously for peripheral occlusive vascular disease presented with three days of red blood per rectum and melanic stools with syncopal events. On initial examination, he was hemodynamically stable, with a soft non-distended abdomen that was tender to palpation in the left lower quadrant. Baseline haemoglobin was 8.1 g/dL, dropping to 6 g/dL four hours after admission. He subsequently became significantly hypotensive and was initially treated with transfusion of four units of packed red blood cells and aggressive intravenous fluid hydration. Contrast-



**Figure 1.** (A) CT scan showing strict contact between the third portion of the duodenum and the proximal edge of the aorto-bifemoral graft with air bubbles extending anteriorly. (B) Gastroduodenoscopy showing a large, full thickness parietal defect >4 cm in diameter involving the entire posterior wall of the third portion of the duodenum with exposure of the prosthetic aortic graft.

enhanced CT revealed a strict contact between the third portion of the duodenum and the proximal edge of the aorto-bifemoral graft, with soft tissue density and air bubbles extending anteriorly (Figure 1(A)). Extravasation of the aortic contrast material into the bowel lumen was noted at the CT scan, confirming the SAEF diagnosis.

### Technique

Endovascular repair was carried out in the emergency setting. Bilateral femoral percutaneous access was gained under local anaesthesia, and an infrarenal bifurcated endovascular stent graft (C3 Excluder; WL Gore & Associates, Flagstaff, AZ, USA) was deployed that extended from the lower edge of the renal arteries down to both iliac branches of the previous aorto-bifemoral prosthesis, excluding the proximal anastomosis and the site of aorto-enteric communication. The endograft was then reinforced at its bifurcation by implanting two balloon-expandable bare metal stents (CP Stent, NuMed, Hopkinton, NY, USA) in “kissing” configuration, to prevent limb compression inside the previous surgical graft. Completion angiography showed good position and patency of the stent graft, with no contrast extravasation or endo-leaks (Figure 2). Blood pressure measurement at the level of the two arterial femoral accesses did not reveal any significant gradient, confirming good patency of both stent graft limbs. In the early post-operative days, the patient showed significant hemodynamic improvement, with no further GI bleeding. Nevertheless, he was still septic with fever and leucocytosis. Microbiological blood and urine cultures were negative, and the broad-spectrum intravenous antibiotics piperacillin/tazobactam and vancomycin were administered. On postoperative day 5, the patient

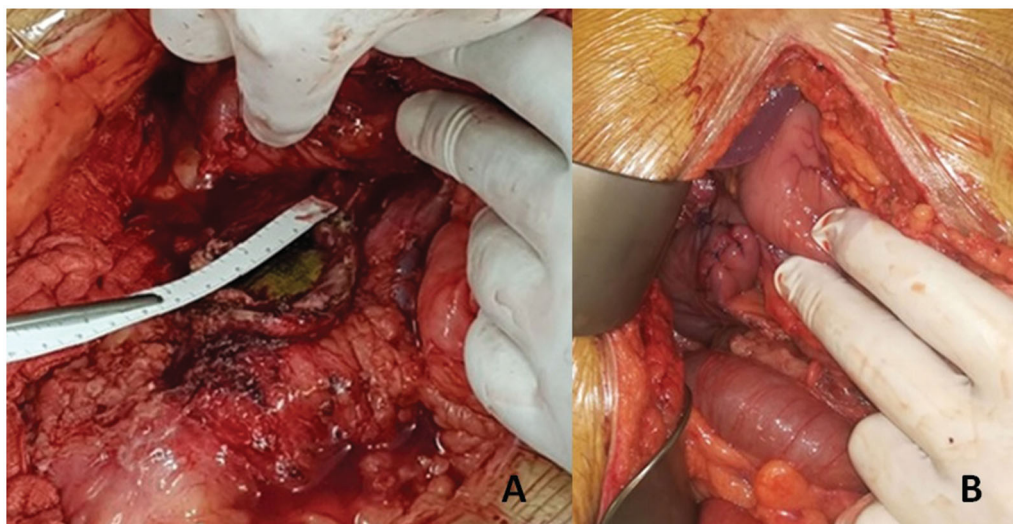
underwent esophagogastroduodenoscopy to assess the extent and location of the duodenal defect. Endoscopy confirmed a large, full-thickness parietal defect >4 cm in diameter involving the entire posterior visceral wall of the third/fourth portion of the duodenum (Figure 1(B)) with exposition of the aortic prosthetic graft.

Ten days after the endovascular procedure the patient was hemodynamically stable with no more signs of sepsis and a second operation was planned to treat the duodenal defect. The extent of this lesion made it impossible to attempt a simple suture of the defect, making duodenal resection necessary (Figure 3(A)). Resection of the third and fourth duodenum was accomplished under endoscopic guidance after extensive adhesiolysis and intestinal de-rotation around the mesenteric axis in a clockwise fashion to achieve mesentery elongation of the proximal jejunum, mandatory for a tension-free latero-lateral duodenojejunal anastomosis fashioned away from the infected field (Figure 3(B)). The complexity of this procedure consisted mainly in the cephalad dissection of the mesenteric root which, in close proximity to the duodenum, looked like it was fused with the viscus and the aortic graft plane. Aorto-bifemoral prosthesis plus endograft radical explant with *in-situ* reconstruction was not attempted at this time due to the excessive surgical impact of such a strategy in a critical condition patient; moreover, no intraoperative evidence of perigraft abscess was found, supporting the rationale of a conservative approach. After peri-aortic debridement completion and thorough rinsing of the field, the aortic prosthetic graft was covered with an omental flap.

After the second surgery, the course was initially uneventful, and early oral re-feeding was started. On postoperative day 3, the patient presented with right



**Figure 2.** Completion angiogram after endograft deployment with stent reinforcement at the level of its bifurcation, showing good patency and no evidence of endoleak.

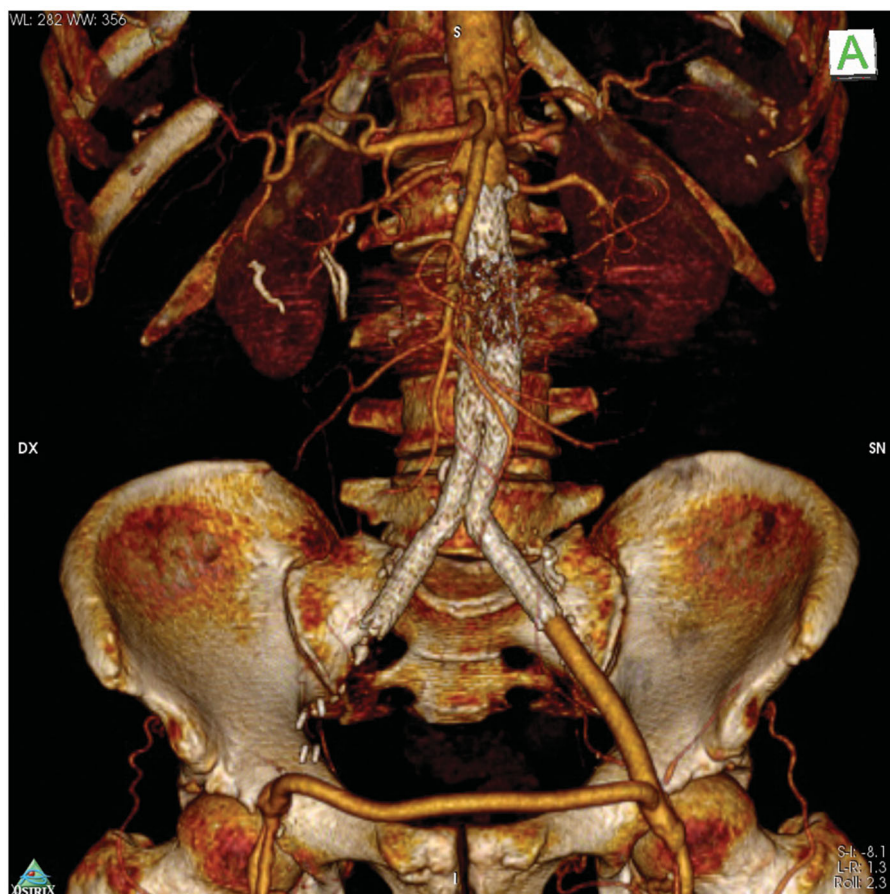


**Figure 3.** (A) After completion of intestinal derotation and resection of the third and fourth portion of the duodenum and the first jejunal loop, the residual fibrotic rim at the site of the aortoenteric fistula was left *in situ* with the aortic graft exposed. After thorough rinsing with saline and local antibiotics, the exposed graft was covered by an omental flap. (B) Latero-lateral duodenojejunal anastomosis fashioned away from the infected field.

lower limb severe ischemia due to thrombotic occlusion of the ipsilateral iliac branch of the endograft. CT-scan imaging did not show any technical defect of stent graft repair that could be responsible for early limb occlusion. The patient underwent emergency femoro-femoral crossover bypass grafting using a 7 mm silver-coated Dacron prosthesis due to its established infection resistance, which restored flow to the affected limb.

Recovery was uneventful until postoperative day 10, when the patient developed a fever with chills. Blood culture showed *Candida albicans*, and caspofungin 50 mg per day was administered in addition to the broad-spectrum antibiotics prescribed after the second surgery, until the blood culture was negative. Ocular fundus examination and echocardiogram, performed to detect possible widespread fungal infection, were negative. CT scan showed no signs of





**Figure 4.** CT-scan at one-year follow-up confirms patency of the left iliac limb of the endograft and of the femoro-femoral crossover bypass, with no sign of residual infection.

perforation or intra-abdominal abscesses and normal flow in the crossover bypass. The patient showed clinical improvement after a few days of antifungal therapy. Caspofungin was continued for 20 days until the blood culture was negative and the patient was discharged six weeks after admission with 400 mg/day oral fluconazole for 30 days.

The one-year follow-up showed no clinical signs of infection and normal laboratory tests including CRP values. CT scan showed good patency of the left limb of the endograft and of the femoro-femoral crossover bypass, and no evidence of graft infection (Figure 4).

## Discussion

Optimal management of SAEF remains unclear. One conventional treatment is extra-anatomic bypass grafting and aortic ligation and subsequent graft removal associated with bowel repair. This technique is associated with a 25–90% operative mortality rate, a 5–25% amputation rate, and a 10–50% aortic stump rupture rate. *In-situ* aortic reconstruction is proposed as a less morbid alternative, but its average perioperative

mortality is 27–30% [5,8,9]. Regardless of the revascularization technique, there is an extremely high mortality rate, mainly due to the technical challenges of the operation, associated anaemia, and septic state at presentation, and the usually poor pre-existing general conditions [10–13]. Recent case reports and reviews describe the endovascular management of aortoenteric fistulae in high-risk patients, which shows lower perioperative morbidity and mortality rates than traditional surgery [2,3,14–20].

In a recent review and pooled data analysis, Kakkos et al. compared different open and endovascular approaches and reported higher in-hospital mortality for open surgery (33.9%, 246/725) versus an endovascular approach (7.1% 7/98,  $p < 0.001$  OR 6.7, 95% CI 3–14.7). However, differences in the mid- and long-term outcomes disappeared during the first 18–24 months after the procedure (two-year survival rates of 51% and 40%, respectively), with a higher sepsis rate for the endovascular approach (two-year rates of 42% vs. 19% for open repair), mainly due to incomplete removal of the infected graft. Persistent or early sepsis during the post-operative period occurred

equally after open and endovascular procedures (52/625, 8.3% vs. 9/95, 9.5%,  $p = 0.71$ ) [21].

In our case, the patient's co-morbidities (severe coronary artery disease) and his poor clinical condition (hemodynamic instability) at admission made an emergency endovascular procedure the best treatment choice to control the damage and allow the patient to recover from life-threatening symptoms. This allowed us to plan an elective open procedure after hemodynamic and septic parameters improved. Conservative treatment was chosen to reduce the risk of post-operative mortality associated with a more extensive procedure including an extra-anatomical bypass grafting that would have more than doubled the operating time.

Moullakis et al. reported a high-risk patient with SAEF who was treated with an endovascular approach plus duodenum restoration without further aortic reconstruction. After 20 months, the patient was symptom-free with normal blood tests and no signs of infection on CT [19]. Tan et al. reported a case of proximal SAEF after open abdominal aortic aneurysm repair that was treated with stent graft and the double-chimney technique in both renal arteries. The second stage of the procedure involved laparotomy to repair the defect in the duodenum to prevent further contamination from the bowel contents [22]. Similar non-curative treatment for SAEF was reported by Hansen et al. who treated a high-risk patient with implantation of an aortic stent graft and fistula revision without original graft removal. The patient survived for 14 years with two recurrences of aortoenteric fistula [23].

In conservative treatment, antibiotic therapy guided by blood, wound, intraoperative drainage, and tissue specimen culture results is essential and should be initiated as soon as SAEF is suspected. Patients usually remain on antibiotic therapy for at least eight weeks; therapy is stopped only when clinical and laboratory markers of an active infectious process are absent.

## Conclusion

SAEF treatment with endovascular exclusion of the aorto-duodenal communication without further aortic reconstruction, followed by resection of the eroded part of the duodenum and appropriate antibiotic therapy shows promise, especially for high-risk patients. Although definitive conclusions cannot be drawn from few cases, this approach seems to reduce the high mortality rate and optimizes postoperative

quality of life. Further evaluation of this technique may be warranted in selected patients to provide a clearer definition of who would require and could benefit from this type of treatment.

## Declaration of interest

No potential conflict of interest was reported by the authors.

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