

Title	Secondary Aortoenteric Erosion Followed by Recurrent Lower Extremity Abscesses
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Journal	Annals of vascular surgery, 42(): 302.e1-302.e5
URL	<a href="http://hdl.handle.net/10130/5048">http://hdl.handle.net/10130/5048</a>
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## **Secondary aortoenteric erosion followed by recurrent lower extremity abscesses**

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## ABSTRACT:

A secondary aortoenteric fistula (SAEF) is a relatively rare complication of aortoiliac reconstructive surgery, often involving relatively fixed duodenal 3<sup>rd</sup> portion and a vascular anastomosis. We observed a 52-year-old man with a recurrent right lower leg abscess following the erosion between ipsilateral bifurcated prosthetic graft limb and non-fixed jejunum. In situ graft-sparing surgical treatment with aggressive debridement was successfully performed. A SAEF may occur even at a non-vascular anastomosis site, or in non-fixed small bowel, and may become a source of a septic embolus. A high index of suspicion for SAEF is required for early diagnosis and treatment of this life-threatening complication.

A secondary aortoenteric fistula (SAEF) is one of the rare complications of abdominal aortoiliac reconstructive surgery, with a reported incidence of 0.36%-3% (1-7). The clinical and radiological manifestations of SAEF are still unspecified, despite improved knowledge regarding the pathology. However, early diagnosis is essential to manage this life-threatening complication, with high operative mortality rates of 40%-50% (8). A fistula may occur when a prosthetic graft erodes into the bowel (a continuous mechanical contact may be necessary for the incidence). The findings from our case highlight the importance of considering the possibility of SAEF in any patient with a history of aortoiliac reconstructive surgery; even appendicitis may cause adhesions between the free bowel and the paraprosthetic site. The patient consented to the publication of this report.

## **CASES REPORT**

A 52-year-old man was admitted to our hospital with a presentation of fever and swelling of right lower leg. Two-and-a-half years prior to admission, he had undergone an emergent bifurcated graft

replacement (Dacron, 18 x 9 mm) for a ruptured abdominal aortic aneurysm (rAAA). Retroperitonization had been performed at the operation. He had been intermittently plagued with high fever and abdominal pain for one year prior to admission. Six-months prior to admission, he developed an abscess in the right lower leg, accompanied by fever. Broad-spectrum antibiotics were administered, followed by open drainage. The blood and pus cultures were negative for growth at that time. The abscess cured promptly, though its etiology was not identified. At the time, he had one episode of hematochezia, although, no specific lesion was detected on colonoscopy.

On the day of admission, his blood pressure was 154/87 mmHg; pulse rate was 67 beats/min; and his temperature was 38.3°C. His abdomen was soft and flat, and his right leg was swollen and reddish. The laboratory findings were as follows: white blood cell count: 13,130/mm<sup>3</sup>, hemoglobin: 12.7 g/dl, and C-reactive protein: 6.93 mg/dL. Enhanced CT scanning revealed air density mass around the prosthetic bifurcated graft, adjacent to the jejunum (Fig. 1A), a phlegmonous appendicitis, and a recurrence of the right lower leg abscess (Fig.1B); however, the blood and

the abscess cultures revealed no growth.

He underwent an open laparotomy through a midline incision. On exploration, the tip of the appendix and the jejunum were found clearly adhered to the swollen retroperitoneum (Fig.2A), however, no other adhesion was apparent. The appendix was slightly swollen without perforation, and the jejunum was eroded by the underlying prosthetic graft limb (Fig.2B). The graft limb was bile-stained, and slightly oozed blood (Fig. 2C). The bile stain was localized on the right limb, and it was completely separated from the vascular suture line. There was only a little pus around the graft.

The bile stained graft limb was resected with a safety margin, which was 3 cm long in total, and anastomosed in an end-to-end fashion. Aggressive debridement of the surrounding tissue was performed. Irrigation with 10,000 mL of normal saline was performed, and no irrigation tube was placed. The jejunum was partially resected and anastomosed in an end-to-end fashion, while an appendectomy was performed at the same time. Using vascularized omentum, the graft was covered and the retroperitoneal space was packed to control infection.

The culture from the resected graft grew *Escherichia coli* and *Enterobacter cloacae*, however, the blood culture remained negative for growth. The mucosa of the resected appendix revealed chronic inflammation. The patient's recovery was uneventful. Based on the cultured organisms, CPFX was administered intravenously for 3 weeks, followed by oral LVFX for 8 months. Blood analysis was performed every 3-month, and an enhanced CT was performed every year, with additional CT scanning if there were any signs of inflammation or infection, and no evidence of re-infection was observed over 5 years after the surgery.

## **DISCUSSION**

A secondary aortoenteric fistula (SAEF) is a rare complication of abdominal aortoiliac reconstructive surgery, which may occur after a few days or up to 27 years after the primary surgery (8-10). The operative mortality is high, reported as 40%-50% (2, 7, 8); thus early diagnosis and aggressive treatment are essential to improve the outcomes. This critical condition often presents with unspecific symptoms, such as fever, abdominal pain, and anorexia (8), which makes it difficult to diagnose

correctly and promptly.

Two types of SAEF have been described (11, 12). Type-1 fistula is a true aortoenteric or graft-enteric fistula, developing between the aortic suture line and the bowel. “Herald bleeding”, which is a minor bleeding preceding major hemorrhage from bowel tract, has been observed and reported in many cases (2, 8). Sepsis and abdominal pain are relatively rare with this type of fistula (11, 12).

Type-2 fistula is a paraprosthetic-enteric erosion, with no communication between the bowel and the graft, as in our case. It is known to account for 15%-20% of SAEF (11, 12). In this type of fistula, the bleeding occurs from the edges of the eroded bowel because of the mechanical pulsations of the aortic graft. Sepsis is more frequently associated with this type of fistula (57%) (11, 12). As a clinical presentation, septic emboli in the lower extremities, septic arthritis, multicentric osteomyelitis and hypertrophic osteoarthropathy have been reported, with an incidence rate of 2%-8% (2, 8, 13-16). An infectious clot may form in an eroding prosthetic graft, and become the source of an embolus.



The etiology of SAEFs still remains unclear, but two possible mechanisms have been reported. One is the constant pulsating contact of the graft on the bowel, and the other is an already existing adhesion due to an infection or inflammation (8, 17, 18). However, SAEFs occur even after endovascular aneurysm repair regardless of the absence of aortic suture line and prosthetic graft outside the aortic lumen, which could make it difficult to understand the exact pathogenesis (19). Most type-1 SAEFs have been reported to involve the relatively fixed duodenal 3<sup>rd</sup> portion and a vascular suture line of the anastomosis (20). In our case, the suture line and the duodenum were not associated with the erosion. The erosion was located between right graft limb and the non-fixed jejunum. We considered the possibility that the chronic appendicitis spread retroperitoneum and caused the adhesions between the jejunum and the lesion, resulting in the origin of constant pulsating contact. It may be possible that the primary surgery for rAAA caused the adhesions between the jejunum and the retroperitoneum, although, the retroperitonization was performed as far as possible at the time of operation. However, that still does not explain why the tip of the appendix penetrated the lesion or

the mucosal inflammation seen in the resected appendix. Moreover, chronic appendicitis would be consistent with the patient's intermittent abdominal pain. Appendicitis involved in SAEF has been reported to be as rare as 3% (8).

Infection control is one of the most important factors in the management of this life-threatening complication. There is no consensus on the optimal duration of antibiotic therapy after the treatment of infected aortic lesions. Parenteral antibiotics are commonly administered for 2 to 8 weeks after surgery; whether lifelong oral antibiotics are needed, is still debated (21-23).

There are several reported operative strategies to treat this severe condition, which are often complex procedures, e.g., graft excision with extra anatomical reconstruction, in situ reconstruction with autogenous vein or cryopreserved allograft or rifampin-soaked grafts, and simultaneous or two-stage surgeries (2, 8, 20, 24, 25). Although extra anatomical reconstruction has improved results, the mortality and aortic stump blowout rate still remain high (26). Replacement of the grafts with biological conduits will most likely remain the best treatment option,

although, there are no universal guidelines on managing prosthetic graft infections (27, 28). Moreover, cryopreserved allograft may not be available for such urgent operations, and it is expensive. Conversely, in situ graft-preserving strategies have also been reported (29-33). Previous reports have described either a prolonged period of irrigation with povidone-iodine, or intermittent packing with povidone-iodine-soaked sponges (32, 33). These techniques require stage-surgical procedures to cover the graft, and povidone-iodine irrigation is limited by its toxicity (29). We resected the bile-stained graft limb with an adequate safety margin and preserved the original graft because we thought the infection was completely localized, and the length of the graft limb was long enough to allow suturing in an end-to-end fashion without a need of even an autogenous vein graft. A single irrigation with normal saline was administered to reduce the residual bacteria count, which was absolutely not toxic and did not require staged therapy to cover the graft.

Furthermore, the omentum has been used for a variety of surgical procedures (34). It can prevent or control aortic graft infection (35), and has been described as an ideal biological drain (30). Our case had a rich

and vascularized greater omentum, and the infection was localized. Our strategy may be a feasible alternative to conventional replacement surgery for such limited cases.

## **CONCLUSION**

The aim of this case report is to emphasize the importance of early diagnosis and treatment of sepsis or lower extremity abscesses in patients who have a history of aortic reconstructive surgery. A SAEF may occur in the prosthetic graft limb even away from the anastomosis, and could be a source of septic embolism. In situ graft-sparing surgical treatment with aggressive debridement may be feasible in the treatment of localized type-2 SAEF.

## **DISCLOSURE**

We all have no conflict of interest to declare.

## **FIGURE CAPTIONS**

Fig.1A The arrowhead shows an air density mass around the prosthetic bifurcated graft limb.

Fig.1B Arrowheads showing the right lower leg abscesses.

Fig.2A The tip of appendix and the jejunum found clearly adhered to swollen retroperitoneum.

Fig.2B The jejunum eroded by the underlying prosthetic graft limb. The arrow shows the jejunal mucosa, and the arrowhead shows the underlying prosthetic graft.

Fig.2C The graft limb is bile-stained and slightly oozing blood. The bile stain is localized, and completely separated from the vascular suture line.

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