Clinical Case Report

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A case report of successful treatment of secondary aortoenteric fistula complicated with gastrointestinal bleeding and retroperitoneal abscess in an elderly patient

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Abstract

Rationale: The treatment of secondary aortoenteric fistula (SAEF) involves maintaining hemodynamic stability, infection control, revascularization, and surgical repair. Conventional open repair is associated with high mortality, whereas endovascular stent-graft repair is associated with recurrent infection or bleeding.

Patient concerns: We report the case of an 85-year-old man with SAEF who presented with gastrointestinal bleeding and retroperitoneal abscess.

Diagnoses: He was misdiagnosed for 5 months. SAEF was eventually diagnosed by CT and gastroduodenoscopy.

Interventions: The patient underwent hybrid open surgery: extraanatomic left axillofemoral bypass graft reconstruction, exploratory laparotomy, aortic stent graft excision, infrarenal abdominal aortic suture, left common iliac artery ligation, extensive surgical debridement, and retroperitoneal abscess resolution and drainage, along with duodenal defect repair and jejunal feeding tube placement.

Outcomes: He survived the complicated surgery and several life-threatening complications with multidisciplinary management. He has kept well for 15 months.

Lessons: Elderly SAEF patients can undergo open repair when circumstances permit, but multidisciplinary management is crucial.

Abbreviations: CT = computed tomography, SAEF = secondary aortoenteric fistula.

Keywords: elderly, gastrointestinal bleeding, retroperitoneal abscess, secondary aortoenteric fistula (SAEF), treatment

1. Introduction

Secondary aortoenteric fistula (SAEF) is a rare yet lethal condition with a 45.8% mortality rate in the first month.[1] SAEF is typically secondary to abdominal aortic aneurysm repair.[2] Early diagnosis is difficult and depends on a heightened clinical suspicion. Uncorrected SAEF is almost always fatal. Clinical outcomes are determined by the timeliness of surgical repair, the revascularization approach (open vs in situ), the type of surgery (emergent vs nonemergent), and the occurrence of complications.[3]

We report a case of SAEF that was successfully treated in an 85-year-old man. Consent for the publication of this case report was obtained from the patient.

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2. Case Report

An 85-year-old man with melena, weakness, and dizziness since 5 months was admitted to our hospital. Thirty-three months ago, he had undergone lower abdominal aorta–common iliac artery bifurcated stent implantation at a local hospital because of a lower abdominal aortic dissecting aneurysm and right common iliac arterial pseudoaneurysm. Twenty-eight months later, he was admitted to the same hospital with melena, tiredness, and dizziness. His hemoglobin level had declined from 11.5 to 7.0 g/dL. Gastroscopy showed superficial gastritis without a definite bleeding source. The gastrointestinal bleeding was attributed to aspirin-induced mucosal injury. Aspirin was stopped, and he was given a blood transfusion and administered proton pump inhibitors, and discharged with a hemoglobin level of 7.8 g/dL.

Two months later, he was admitted to the same hospital with complaints of progressive fatigue, loss of appetite, weight loss, intermittent palpitation, and intermittent fever. On this occasion, both leukocytosis and anemia were detected (white blood cells, 15,450/μL; hemoglobin, 7.8 g/dL). He was diagnosed with pneumonia and prescribed antibiotics. He was discharged 18 days later without significant improvement.

Two weeks before admission to our hospital, in addition to the above symptoms, he began experiencing right back pain, right knee movement restriction, and right leg swelling. On physical examination, he appeared pale and skinny. His vital signs were as follows: body temperature, 38.2°C; pulse rate, 96/min, regular; respiratory rate, 18/min; and blood pressure, 120/96 mm Hg. His abdomen was soft and nontender, with no palpable masses and a

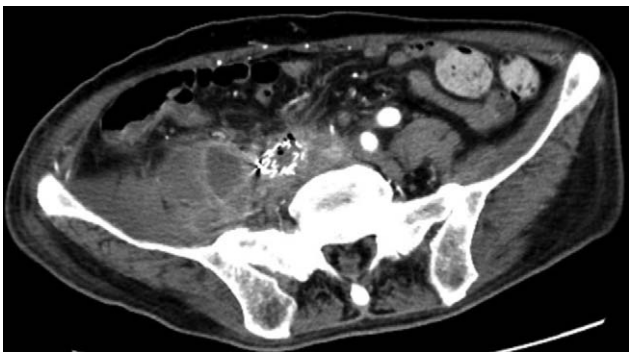

Figure 1. Computed tomography scan showing a gas shadow in the right external iliac artery and encapsulated fluid around the right psoas major and iliacus muscles.

normal frequency of bowel sounds. He had severe anemia (hemoglobin, 5.8 g/dL). His white blood cell count was 8130/mm^3 (82.3% granulocytes). He was given a blood transfusion and administered antibiotics. Computed tomography (CT) showed a gas shadow in the right external iliac artery and around the lower abdominal aorta and common iliac artery (Fig. 1) and thrombosis in the right common iliac artery, right external iliac artery (Fig. 2), and right common iliac vein. Encapsulated fluid was observed around the right psoas (Fig. 1). SAEF was diagnosed. Duodenoscopy revealed a fistula in the third part of the duodenum as the source of bleeding.

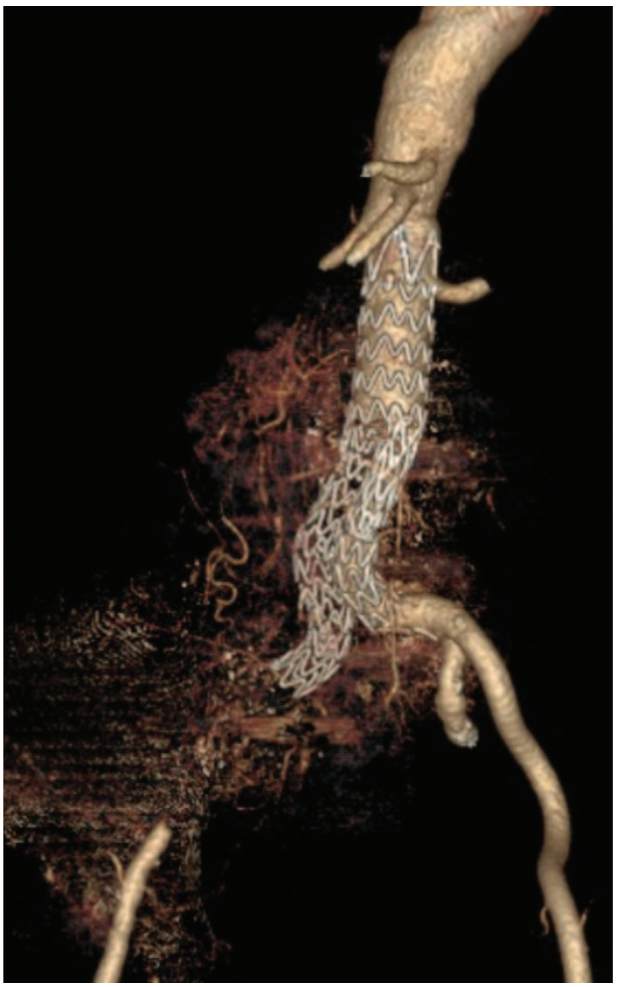

Figure 2. Three-dimensional view of the thrombus in the right common iliac artery and right external iliac artery.

Based on the patient's clinical features and CT findings, the fistula was considered to be related to stent-induced infection and intestinal erosion. We elected to perform open surgery with extraanatomic bypass in this patient, as this enables the safe excision of the infected endograft, eradicating the source of infection. In contrast, endovascular repair involves in situ graft replacement and partial or complete graft excision. The patient underwent hybrid open surgery: extraanatomic left axillofemoral bypass graft reconstruction, exploratory laparotomy, aortic stent graft excision (Fig. 3A), infrarenal abdominal aortic suture, left common iliac artery ligation, extensive surgical debridement, and retroperitoneal abscess resolution and drainage (performed by vascular surgeons), along with duodenal defect repair and jejunal feeding tube placement (Fig. 3B). The operation time was 7 hours. The patient was transferred to the intensive care unit and administered broad-spectrum intravenous antibiotics (imipenem/cilastatin) under multiorgan function monitoring. Tissue from the retroperitoneal abscess grew *Enterococcus faecium* and *Candida albicans*. Therefore, vancomycin and fluconazole were administered.

After 10 days, the patient was transferred to the ward, where he developed hospital-acquired pneumonia, bilateral pleural effusion, and heart failure. He was prescribed wide-spectrum intravenous antibiotics (imipenem/cilastatin and later cefoperazone/sulbactam) and fluconazole. His fluid intake was restricted. His condition gradually improved, and antibiotic administration was terminated. However, he developed progressive abdominal distension. Plain abdominal radiography revealed partial small-bowel obstruction possibly caused by postoperative intestinal adhesions. This complication was managed with fasting, parenteral nutrition, gastrointestinal decompression, inhibition of gastric acid secretion, oral paraffin fluid, and enema. He recovered and has kept well without recurrent infection or anemia for 15 months now.

3. Discussion

The etiopathogenesis of SAEF involves several mechanisms[4]: bacterial seeding of a prosthetic graft, allowing extensive infection to the suture line and resulting in anastomotic failure, perigraft infection, and SAEF formation; bowel damage, especially during emergency graft insertion, causing bowel-wall trauma and ischemia; mechanical injury, including devascularization, trauma, suture-line failure, and graft inflammation; mechanical factors related to continuous pulsatile movement between the graft and the intestines; pseudoaneurysm or paragraft abscesses compressing, eroding, or invading the intestinal lumen; and mechanical erosion of the intestinal wall owing to graft-induced infection and inflammation.

The commonest symptom of SAEF is gastrointestinal bleeding (e.g., melena, herald bleeding, coffee ground emesis, bright red blood per rectum, severe hemorrhagic shock).[1,5,6] SAEF is difficult to diagnose because of its rarity. In our patient, the cause of gastrointestinal bleeding, which manifested as melena, chronic fatigue, dizziness, and anemia, was misdiagnosed for 5 months. Therefore, we consider that SAEF should be routinely ruled out in patients who present with gastrointestinal bleeding after abdominal aortic aneurysm repair.

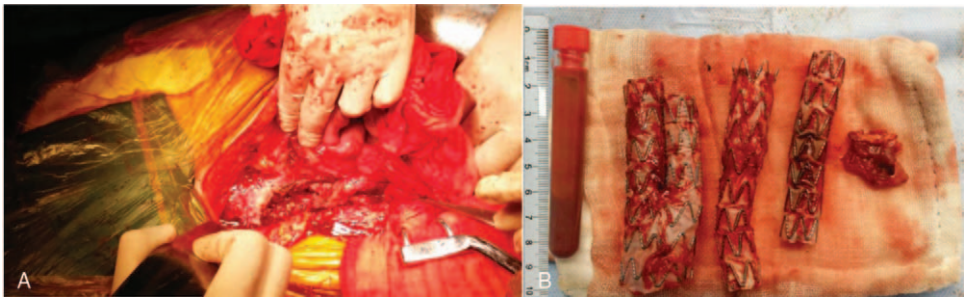

Figure 3. (A) Secondary aortoenteric fistula and (B) excised aortic stent graft.

Other symptoms of SAEF are fever and sepsis. Our patient had intermittent fever with leukocytosis for >3 months, which was misdiagnosed as pneumonia. Other rare symptoms of SAEF include pseudoaneurysm, retroperitoneal abscess, pulsating abdominal mass, groin mass, limb ischemia, abdominal pain, back pain, and weight loss.[6,7] Our patient presented with anorexia, malaise, weight loss, intermittent palpitation, back pain, and right knee movement restriction due to a right psoas abscess.

CT and gastroduodenoscopy are frequently used to diagnose SAEF. The diagnostic sensitivity of CT for SAEF is 92% to 94%.[6,8,9] The most characteristic CT finding is a gas shadow in or around an endovascular graft (Fig. 1), which has 40% diagnostic sensitivity and 100% specificity.[10] Other characteristic CT findings include pseudoaneurysm, visible graft, swelling or hematoma around the graft, soft-tissue mass (measuring >5 mm) around the aorta, intravenous contrast within the gastrointestinal lumen or around the aorta, and duodenal hematoma.[10–12] Additionally, CT can be used to identify infection or abscess if present.[13] Gastroduodenoscopy is recommended in suspected SAEF patients with upper gastrointestinal bleeding to exclude other causes of bleeding. Since most SAEFs occur in the third or fourth part of the duodenum, duodenoscopy or enteroscopy may be necessary to visualize the distal duodenum and proximal jejunum.[14,15]

SAEF management should include the early administration of broad-spectrum antibiotics covering gram-positive and gram-negative organisms and anaerobes.[7,13] The infectious focus should be surgically removed. Failure to control sepsis may result in mortality rates of 60%.[16] Antibiotics should be adjusted according to the results of culture and sensitivity tests of blood, intraoperative drainage fluid, and tissue specimens. Our patient was administered broad-spectrum antibiotics perioperatively, and received vancomycin and fluconazole after tissue from the psoas abscess grew *E faecium* and *C albicans*. Aortic graft infection secondary to *Candida* species is unusual, with systemic *Candida* infections commonly occurring in immunocompromised or debilitated patients.[17] In our patient, the infection was successfully controlled by complete removal of the infectious focus and prompt antifungal and antibiotic therapy.

Conventional SAEF treatment consists of graft removal and extraanatomic bypass, which require a long operation time and pose a high risk of complications associated with aortic clamping, such as thrombosis, infection, and decreased perfusion of the colon and lower limbs.[18] In contrast, endovascular in situ graft replacement and excision shortens the operation time but still carries a high risk of infection and recurrent bleeding.[16]

The prognosis of patients with SAEF may depend on their hemodynamic status at presentation, the timeliness of surgery, the operative technique performed, and the time to surgical exploration.[2] Preoperative shock was associated with postoperative mortality. A delay in surgical exploration may increase mortality, and an uncorrected SAEF is almost always fatal. In one report, early perioperative mortality after the surgical management of SAEF was 45.8%.[1] In a recent report, the 1-month, 1-year, and 5-year mortality rates of SAEF were 25%, 30%, and 34%, respectively.[7] Optimal outcomes of SAEF depend on maintaining hemodynamic stability, infection control via empiric intravenous antibiotics, revascularization, surgical repair of the underlying defect, and maintenance of blood perfusion to the lower limb.[10,16] Currently, there are no established guidelines for the repair of the SAEF. Our patient underwent nonemergent open surgery to eradicate the infectious source and repair the duodenal defect. Although he developed several life-threatening complications, he recovered under the supervision of a multidisciplinary management team that included vascular surgeons, gastrointestinal surgeons, intensivists, and internists. He has been well for 15 months now. In elderly and debilitated SAEF patients, multidisciplinary management is essential.

4. Conclusions

The diagnosis of SAEF, a rare, life-threatening complication of abdominal aortic aneurysm repair, depends on clinical, CT, and gastroduodenoscopy findings. Early diagnosis is difficult and remains dependent on a heightened clinical suspicion. The surgical approach (open vs endovascular) for SAEF repair should be individualized. In elderly patients, multidisciplinary management is crucial.

Author contributions

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Supervision: Hongwei Li, Xueming Chen, Jianshe Li.
Writing – original draft: Chunyan Jiang.
Writing – review & editing: Hongwei Li, Chunyan Jiang.

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