

# Polymicrobial Bacteremia with Insidious Gastrointestinal Blood Loss in A Patient with A Remote History of Aortobifemoral Bypass: A Rare Diagnosis of Secondary Aortoenteric Fistula

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## Abstract

Secondary aortoenteric fistula is a rare yet fatal complication after reconstructive surgery of aortic aneurysm. A 68-year-old man with a significant past medical history of type 2 diabetes, coronary artery disease status post cardiac stent, and aortobifemoral bypass graft performed 10 years ago, presented with shortness of breath, chills, and progressive anemia for the past 14 months. Patient reported one episode of melena 2 weeks before admission. The initial blood cultures growth of a variety of microorganisms (*Streptococcus sanguis*, *Lactobacillus paracasei*, *Candida lusitanae*) and the subsequent blood cultures growth of another microorganism (*Enterobacter Cloacae*) were considered to have a GI source. A contrast CT scan of abdomen and pelvis did not show acute intra-abdominal process. Then a FDG PET-CT scan result showed hypermetabolism signal associated with the proximal end of the aortoiliac graft, concerning for infection of the graft itself. An esophagogastroduodenoscopy revealed a large centrally bulging lesion, without bleeding, in the third part of the duodenum, which was consistent with a secondary aorto-enteric fistula formed by infected aortic graft eroding into duodenum. The patient subsequently underwent explantation of the infected aortobifemoral bypass graft, reconstruction with cryopreserved aortoiliac allograft, and resection of small intestine with anastomosis closure of duodenotomy. The patient was discharged from the hospital 10 days after the operation and later was doing well. Secondary AEF should be suspected in patients presenting with unclear source of bacteremia with or without GI bleeding and a history of aortic repair. Clinical suspicion is the most crucial factor contributing to the right diagnosis. (J Intern Med Taiwan 2022; 33: 378-382)

**Key Words:** Secondary aortoenteric fistula

## Introduction

Aortoenteric fistula (AEF) is an abnormal connection between the aorta with the gastrointestinal tract. AEF occurring in the setting of the native aorta, or a previously untreated aneurysm is con-

sidered as a primary fistula, while those that occur following aneurysm repair (e.g., endovascular aneurysm repair or EVAR) are termed secondary fistula. This risk of secondary AEF after EVAR was rare, 0.4 to 1.6%, but associated with significant morbidity and mortality<sup>1,2</sup>.

Secondary AEF after EVAR often present with nonspecific vague symptoms, including fever, abdominal pain, loss of appetite and weight, and low-grade infection<sup>3,4</sup>. Clinically, secondary AEF present in a variety of ways, which makes a prompt diagnosis challenging and relying on high clinical suspicion from the clinicians. We present a patient with progressive anemia, gastrointestinal blood loss, and recurrent polymicrobial bacteremia before being diagnosed.

## Case report

A 68-year-old man with a significant past medical history of type 2 diabetes, COPD, coronary artery disease status post cardiac stent, and aortobifemoral bypass graft performed 10 years ago, was admitted with presentations of shortness of breath, chills but no fever, and progressive anemia for the past 14 months. ECG and trend of cardiac enzymes were negative. Chest CT imaging revealed no evidence of pulmonary embolism. He was hemodynamically stable. The pertinent initial labs showed a hemoglobin of 7.5 g/dl (14 months earlier 11.9 g/dl) and elevated inflammatory markers (leukocytes 9.8/nL, CRP 85.3 mg/dL) with transient lactic acidosis. Blood cultures on admission showed positive of a variety of microorganisms (*Streptococcus sanguis*, *Lactobacillus paracasei*, *Candida lusitaniae*). Patient reported one episode of red blood per rectum and melena between 1 to 3 weeks before admission.

The initial blood cultures growth of a variety of microorganisms (*Streptococcus sanguis*, *Lactobacillus paracasei*, *Candida lusitaniae*) and the subsequent blood cultures growth of another microorganism (*Enterobacter Cloacae*) were considered to have a GI source. A contrast CT scan of abdomen and pelvis did not show acute intra-abdominal process. Then a FDG PET-CT scan result showed hypermetabolism signal associated with the proximal end of the aortoiliac graft, concerning for infection of the graft itself (Figure 1). A diagnos-

tic esophagogastroduodenoscopy (EGD) revealed a large centrally bulging lesion, without bleeding, in the third part of the duodenum (Figure 2a) and close-up view suggested of foreign body like texture of material (Figure 2b). The EGD finding in the third part of duodenum was consistent with a secondary aorto-enteric fistula formed by infected aortic graft eroding into duodenum and served as the source of recurrent polymicrobial bacteremia. The patient subsequently underwent explantation of the infected aortobifemoral bypass graft (old graft with fistula to the duodenum), reconstruction with cryopreserved aortoiliac allograft, and resection of small intestine with anastomosis closure of duodenotomy. The removed aortic graft cultures grew a variety of microorganism (*Enterococcus faecalis*, *Enterobacter Cloacae*, *Candida lusitaniae*). The patient was discharged from the hospital 10 days after the operation and when examined 2 months later he was doing well.

## Discussion

AEF is a rare but devastating diagnosis with gastrointestinal hemorrhage, which can range from insidious blood loss to exsanguinating hemorrhage, and infections as the most common initial presentations. Interestingly, a landmark multicenter study on aortoenteric fistulization after stent grafting of

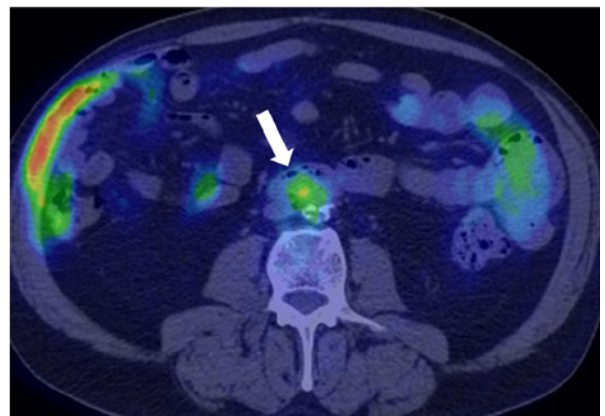


Figure 1. A signal of increased metabolism associated with the proximal end of the aortoiliac graft, concerning for infection of the graft itself.



Figure 2. (a) A large excavated and diverticular-like lesion, without bleeding, in the third part of the duodenum. (b) A close-up view suggested of foreign body like texture of material.

the abdominal aorta (MAEFISTO) reported less than <20% of patients presenting with hemorrhagic shock as the aortic aneurysm is typically excluded by the deployed stent graft<sup>5</sup>. The interval between aortic reconstruction and the development of secondary AEF can range from a few months to several years (median of 32 months)<sup>5</sup>. Seventy-five percent of secondary AEFs are located between the distal duodenum and the proximal suture line of the aorta<sup>6</sup>. Another case series reported 40% of fistulas located in the duodenum, 40% in other areas of small intestine and 20% in colon<sup>7</sup>.

Mechanisms for primary AEF include atherosclerosis (60-85%) and infection (15%), and less commonly are a result of tumors, duodenal ulcers, foreign materials or complications after radio- or thermal therapy<sup>8,9</sup>. For secondary AEF, at least two hypotheses have been postulated. One suggests a continuous mechanical stimulation due to aortic pulsation directly affecting the intestinal walls. The precipitating factors include stent migration, stent fracture or kink, suture line failure, adhesion between aorta and bowel wall, and pseudoaneurysms which may erode the bowel lumen<sup>10</sup>. The observation of most secondary AEFs affecting the third or fourth portion of the duodenum, which are compressed between the superior mesenteric artery

and abdominal aorta in the retroperitoneal space, further support this hypothesis<sup>11</sup>. The other hypothesis suggests an origin in a local inflammatory process and periaortic tissue necrosis due to prosthesis infection<sup>12</sup>. Periaortic inflammation appeared stronger in patients with previous aortic surgical repair than in patients with only previous EVAR. The observation of significantly higher secondary AEF (3.9%) in patients with prior surgical repair of aneurysm than in patients with only atherosclerotic aortic aneurysm (<0.5%) in the MAEFISTO multicenter study further supports this hypothesis<sup>5</sup>.

Contrast CT is typically the first imaging study on patients who are suspected with AEF, and it makes the diagnosis in 33-80% of the patients<sup>4,13</sup>. Characteristic CT findings include the presence of gas around the graft and periaortic fluid collections. FDG-PET scans can be used to detect prosthetic graft infection.

The diagnostic sensitivity of EGD for secondary AEF was reportedly only 24%<sup>6</sup>. This is related to the following factors: variable operative experience, gastroscopy only reaching the second segment of the duodenum and missing the third and fourth segments which are the most AEFs located, and the endoscopist's bias of misdiagnosing the endoscopic findings of AEF as ulcers, polyps, erosion<sup>7</sup>. Sug-

gestive endoscopic findings, including ulceration, excavated lesion or foreign body like material, submucosal tumor with a small ulcer, or active hemorrhage, recognized by the endoscopist carefully examining up to the fourth part of duodenum contribute to the right diagnosis<sup>14</sup>. Interestingly, in the literature review we found a case report of secondary AEF with the nearly identical and fascinating endoscopic appearance as ours<sup>15</sup>. In the scenario of obscure GI blood loss of AEF patients, CT angiography could be of limited value and the capsule endoscopy can play a valuable role<sup>16</sup>.

Once a secondary AEF is diagnosed, the patient needs surgical intervention as soon as possible, since mortality rate is nearly 100% if untreated<sup>17</sup>. The common surgical approach involves the debridement of the infected tissues, removal of the graft and revascularization, and repair of the enteric defect. For AEF patients treated surgically, including aortic stent graft explantation, in situ aortic reconstruction, and extra-anatomic bypass, the perioperative mortality was reported up to 37% in a landmark multi-center study<sup>5</sup>.

In conclusion, a secondary AEF should be suspected in patients presenting with unclear source of bacteremia with or without GI bleeding and a history of aortic repair. High clinical suspicion is the most crucial factor contributing to the right diagnosis.

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# 多菌叢菌血症合併慢性消化道出血在一位有過主動脈 雙股動脈搭橋手術的病患：一個主動脈腸道瘻管的 罕見案例

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## 摘 要

次發性主動脈腸道瘻管是在病患接受過主動脈瘤重建修復手術後相當罕見但死亡率高的嚴重併發症。我們在這裡報告一位68歲男性病患，因為呼吸急促，慢性貧血，發燒寒意住院。過去病史包括糖尿病，冠狀動脈疾病，而且在十年前接受過主動脈瘤重建修復手術。病患在住院二周前曾經有過黑便。剛住院時的血液培養報告發現多菌叢菌血症(*Streptococcus sanguis*, *Lactobacillus paracasei*, *Candida lusitanae*)，後續追蹤血液培養又發現另一種細菌(*Enterobacter Cloacae*)。這些血液培養發現的菌叢跟消化道菌叢似乎有相關性。為了找出感染源，正電子發射斷層掃描(FDG PET-CT)發現在腹部主動脈移植物上端接近十二指腸位置，有異常代謝亢進訊號，懷疑似感染源。胃鏡檢查發現在十二指腸第三段位置，有一大型憩室樣異常結構，並有異物質地隆起，但並無出血現象，經仔細判讀確認是次發性主動脈十二指腸腸道瘻管。因此，病患接受手術移除已經損壞並感染的主動脈移植物，重新修復主動脈瘤重建手術，並修補十二指腸腸道瘻管，病患術後恢復順利出院。如果病患過去病史有接受過主動脈瘤重建修復手術，發生不明原因感染菌血症，無論是否臨床上有無明顯消化道出血現象，次發性主動脈腸道瘻管一定要列入鑑別診斷來考慮，保持高度警覺和懷疑，是臨床上迅速並正確診斷的基本的要件。