Case Report

Mycotic Iliac Artery Aneurysm Caused by *Clostridium difficile* in a Patient with Axillobifemoral Bypass for Leriche Syndrome

Katsuaki Tsukioka, MD, PhD,1 Kohei Takahashi, MD, PhD,1 Toshihito Gomibuchi, MD,1 Tetsuya Kono, MD, PhD,1 Takahiro Yajima, MD, PhD,2 and Mafumi Owa, MD, PhD2

A 74-year-old man on hemodialysis developed a mycotic aneurysm caused by *Clostridium difficile*. To the best of our knowledge, this is only the second case of such an aneurysm reported in the literature. He had previously undergone axillobifemoral bypass grafting because of symptomatic infrarenal aortic stenosis. Although no blood flow was detected in his occluded right common iliac artery, it expanded rapidly despite intensive antibiotic therapy. As the blood supply to the lower limbs was already secured, only resection of the infected arteries was performed.

**Keywords:** *Clostridium difficile*, mycotic aneurysm, Leriche syndrome

**INTRODUCTION**

Extra-intestinal *Clostridium difficile* infection is rare. Although *Clostridium septicum* aortitis has occasionally been reported in patients with gastrointestinal malignancy,1,2) to the best of our knowledge there is only one previously reported case of mycotic aneurysm caused by *C. difficile*.3) We present a patient with *C. difficile* aortitis who developed a rapidly expanding mycotic aneurysm of the right common iliac artery, which was occluded and had previously been bypassed with an axillobifemoral graft because of symptomatic Leriche syndrome. The mycotic aneurysm was surgically resected.

**CASE REPORT**

A 74-year-old man with dialysis-dependent chronic renal failure was admitted to our hospital with dyspnea and septic shock due to pneumonia. He had undergone axillobifemoral bypass grafting because of symptomatic occlusion of the infrarenal abdominal aorta (Leriche syndrome) 3 years previously. Twenty-two days before admission, he had undergone surgery for appendicitis with peritonitis. He had an uneventful recovery and had been discharged 7 days later.

Examination at the time of admission showed blood pressure 90/50 mmHg, pulse rate 74 beats/min, and temperature 39.0°C. Laboratory testing showed a decreased neutrophil count of 2830/mm3 and an elevated C-reactive protein (CRP) level of 25.0 mg/dL (normal range ≤0.3 mg/dL). He was treated with intravenous meropenem (0.5 g/day for 2 weeks) and his sepsis resolved. *Pseudomonas aeruginosa* was grown from blood and sputum cultures. He developed persistent diarrhea, and colonoscopy on day 15 after admission showed pseudomembranous colitis. Stool immunoassay for *C. difficile* toxin was negative. He was discharged on day 45 after admission.

Twenty-eight days later, he was readmitted because of general fatigue. He had a temperature of 39.1°C but...
artery. Extensive debridement was performed, with excision of the infrarenal abdominal aorta and both common iliac arteries. The resected arteries had thickened walls and were filled with fragile thrombi (Fig. 2). The aortic stump was reinforced with pieces of resected parietal peritoneum (black arrow).

On day 5 after aortic surgery, we performed reoperation because of a gradually expanding retroperitoneal hematoma in the left lateral abdominal wall caused by bleeding of the left inferior epigastric artery, which had no abdominal or back pain. His blood pressure was 160/80 mmHg and his pulse rate was 63 beats/min. Laboratory testing showed a normal neutrophil count of 7100/mm³ and an elevated CRP level of 8.8 mg/dL. Computed tomography (CT) revealed expansion of his occluded right common iliac artery to 22 mm in diameter, with surrounding edema (Fig. 1a). As these findings were consistent with a diagnosis of mycotic aneurysm, empirical antibiotic therapy was started with intravenous meropenem (1 g/day) and vancomycin (0.5 g/day). Blood cultures grew no pathogens, but his mild fever continued and his CRP level remained slightly elevated at 4.8 mg/dL on day 6 after readmission. Repeat CT on day 6 after readmission showed an increase in the diameter of the right common iliac artery to 26 mm. Repeat CT on day 18 after readmission showed further expansion of the right common iliac artery to 29 mm (Fig. 1b). We considered that antibiotic therapy would not resolve his infection, and planned surgical resection the following day.

Laparotomy through a midline incision revealed a thickened retroperitoneum with inflammatory changes, but no pus, and an aneurysm of the right common iliac artery. Extensive debridement was performed, with excision of the infrarenal abdominal aorta and both common iliac arteries. The resected arteries had thickened walls and were filled with fragile thrombi (Fig. 2). The aortic stump was reinforced with pieces of resected parietal peritoneum (open arrow). The aortic stump was reinforced with pieces of resected parietal peritoneum (black arrow).

Fig. 1 (a) Enhanced computed tomography (CT) images at the time of readmission showing an occluded right iliac artery, expanded to 22 mm in diameter, with surrounding edema (white arrow). (b) CT image on day 18 after readmission, showing further expansion of the right iliac artery to 29 mm in diameter, with surrounding edema (black arrow).

Fig. 2 Surgical view showing the aneurysmal right common iliac artery. The resected arteries had thickened walls and were filled with fragile thrombi (open arrow). The aortic stump was reinforced with pieces of resected parietal peritoneum (black arrow).

Fig. 3 Histological examination showing granulation tissue with inflammatory cell infiltration and Gram-positive rods (white arrows) (hematoxylin and eosin stain, × 200).
been injured during resection of the parietal peritoneum. The patient then recovered slowly but uneventfully and was discharged to another hospital for rehabilitation on day 38 after aortic surgery. The cultured *C. difficile* was sensitive to ampicillin/sulbactam, vancomycin, and minomycin. He was treated with intravenous ampicillin/sulbactam for 3 weeks, followed by oral metronidazole.

**DISCUSSION**

Mycotic aneurysms have been reported to account for 1.3% of aortic and iliac artery aneurysms. Such aneurysms are most commonly caused by *Staphylococcus* and *Salmonella* spp but may also be caused by anaerobic bacteria. Myotic aneurysms caused by *Clostridium* spp are rare, and to the best of our knowledge, only one myotic aneurysm caused by *C. difficile* has previously been reported, although *C. septicum* aortitis has occasionally been reported. A review by Seder et al. found that 91% of patients with *C. septicum* arteritis also had colonic adenocarcinoma or polyps. They suggested that an anaerobic environment in such rapidly growing tumors is suitable for growth of *Clostridium* spp and bacteria then disseminate into the systemic circulation and infect atherosclerotic lesions, causing myotic aneurysms. Although little is known about the pathophysiological mechanisms associated with *C. difficile* arteritis because of its infrequent occurrence, similarities can be expected with *C. septicum* arteritis. *C. difficile* toxins might, therefore, cause mucosal damage resulting in bacteremia. A review by Libby et al. found that all adult patients with *C. difficile* bacteremia had concomitant gastrointestinal pathology such as diarrhea, small bowel obstruction, or ischemic colitis, which may compromise the integrity of the gastrointestinal tract. They reported that *C. difficile* bacteremia was strongly associated with prior antibiotic exposure but that there was no significant association between *C. difficile* arteritis and colonic tumors. Jacobs et al. reviewed reported extracolonic *C. difficile* infections such as osteomyelitis, visceral abscesses, and prosthetic joint infections, and found that transitory *C. difficile* bacteremia was implicated as a cause of these infections. In our patient, colonoscopy performed after intensive antibiotic therapy for septic shock showed pseudomembranous colitis, which is well known to be caused by *C. difficile* infection, but stool toxin testing and blood cultures were negative. García-Lechuz et al. reported that extracolonic *C. difficile* infection does not always produce toxins, and that blood cultures are rarely positive. In our patient, the atherosclerotic lesions in the occluded arteries, where oxygen delivery and pH were reduced, provided an environment where *C. difficile* was able to grow and form a myotic aneurysm. Libby et al. reported that compromised immunological status was associated with extracolonic *C. difficile* infection. In our patient, chronic renal failure may have contributed to providing suitable conditions for *C. difficile* to infect the atherosclerotic lesions.

The 6-month overall mortality rate in patients with myotic aneurysms caused by *C. septicum* has been reported to be 64% and 100% in those who did not undergo surgery. In our patient, the myotic aneurysm formed in a chronically occluded common iliac artery where blood flow was minimal. Contrary to our expectation that expansion of the aneurysm would be controlled by broad-spectrum antibiotics because the intra-arterial pressure was low, the artery increased rapidly in diameter. Postoperative mortality after myotic aneurysm repair is 33%–63% if the aneurysm has ruptured, compared with 0%–13% if the aneurysm has not ruptured. It has, therefore, been suggested that antibiotic therapy alone is insufficient, and surgical treatment is indicated to avoid rupture.

The *C. difficile* isolated from our patient’s myotic aneurysm was sensitive to metronidazole and ampicillin/sulbactam. The optimal duration of intensive antibiotic therapy in cases of *C. difficile* bacteremia or *C. septicum* aortitis has not been established. As infection may occasionally recur, postoperative antibiotic therapy in patients with *C. septicum* aortitis should be administered for at least 6–8 weeks as recommended for other myotic aneurysms, should probably be continued for at least 3 months, and not be discontinued until no further signs of infection are detected.

**CONCLUSION**

Mycotic aneurysms caused by *C. difficile* are rare but may be encountered after gastrointestinal inflammation in patients with atherosclerotic lesions. In our case, bacteremia secondary to pseudomembranous colitis might have caused infection of the occluded arteries, which subsequently developed unexpected expansion in spite of intensive antibiotic therapy.

**DISCLOSURE STATEMENT**

The authors have no conflicts of interest to declare.
REFERENCES


