

➤ **Case Report** ◀

Two Cases of Refractory Diarrhea Subsequent to Vascular Reconstruction for Infective Superior Mesenteric Artery Aneurysms

Taiki Niki , MD, Naoto Fukunaga, MD, Tatsuto Wakami, MD, Akio Shimoji, MD, Kosuke Yoshizawa, MD, and Nobushige Tamura, MD

Superior mesenteric artery (SMA) aneurysm and their surgical interventions are rare, leading to infrequent reports of postoperative complications. This report describes 2 cases of refractory diarrhea following vascular reconstruction for infectious SMA aneurysms. Both patients underwent aneurysm resection and SMA reconstruction but experienced persistent diarrhea despite treatment with anti-diarrheal medications. Postoperative diarrhea, a complication observed after resection of the nerve plexus around the SMA in gastrointestinal surgery, may be attributed to intraoperative injury to the nerve plexus in our cases. Though palliative therapy is partially effective, more efficacious management strategies are desirable to address this persistent complication.

Keywords: superior mesenteric artery aneurysm, postoperative diarrhea, nerve plexus

Introduction

Superior mesenteric artery (SMA) aneurysms are uncommon vascular pathologies, leading to infrequent reports of postoperative complications following surgical intervention.

This report aims to discuss the possible managements of refractory diarrhea subsequent to infective SMA aneurysm resection and vascular reconstruction.

Case Report

Case 1: A 79-year-old patient presented with acute abdominal pain and fever. His past medical history included


Department of Cardiovascular Surgery, Hyogo Prefectural Amagasaki General Medical Center, Amagasaki, Hyogo, Japan

Received: July 21, 2024; Accepted: September 4, 2024

Corresponding author: Taiki Niki, MD. Department of Cardiovascular Surgery, Hyogo Prefectural Amagasaki General Medical Center, 2-17-77, Higashinaniwa-cho, Amagasaki, Hyogo 660-8550, Japan

Tel: +81-80-2466-2043

E-mail: taikitwotree2481@gmail.com

 This work is licensed under the Creative Commons Attribution-NonCommercial-ShareAlike International license.
©2024 The Editorial Committee of Annals of Vascular Diseases.

aortic stenosis and regurgitation, atrial fibrillation, gastric and esophageal cancer, and chronic kidney disease (CKD). Computed tomography angiography (CTA) confirmed SMA embolism due to atrial fibrillation, leading to emergent thrombus aspiration and stent placement. However, 2 days later, *Staphylococcus epidermidis* was detected in the blood culture taken during the admission fever workup, and further investigation, including ultrasonographic cardiography, revealed that the embolism originated from infective endocarditis affecting the aortic valve. Despite initial antibiotic therapy for 6 weeks, aortic regurgitation progressed, and CTA identified an SMA pseudoaneurysm with an abscess around the stent (Fig. 1A).

The simultaneous intervention for both aortic valve and SMA was considered excessively invasive. Therefore, initially, the patient underwent aortic valve replacement (AVR) and repair of a sinus of Valsalva perforation. Following a 3-month period, with the improvement of his general condition, the resection of the infective SMA aneurysm and reconstruction using an expanded polytetrafluoroethylene graft were conducted (Fig. 1B). Extensive intraabdominal adhesion resulting from previous gastrectomy and esophagectomy complicated identification and exposure of SMA. Consequently, the adventitia of SMA was exposed extensively, spanning from the root to its branches. The postoperative course was uneventful, and the patient was discharged on the 15th day after the second surgery.

At a 3-month outpatient visit, the patient reported persistent diarrhea. Despite thorough examinations, including laboratory analysis, colonoscopy, and stool culture, no specific cause was identified. Since the organic pathologies were ruled out, loperamide was prescribed as palliative therapy. While this reduced the frequency of diarrhea from 10 to 15 episodes per day to 5, complete resolution was not achieved. Notably, the patient adopted a cautious approach, limiting oral intake in an attempt to mitigate diarrhea, resulting in frailty and renal impairment. The patient died due to uremia with worsening CKD approximately 1 year post-surgery.

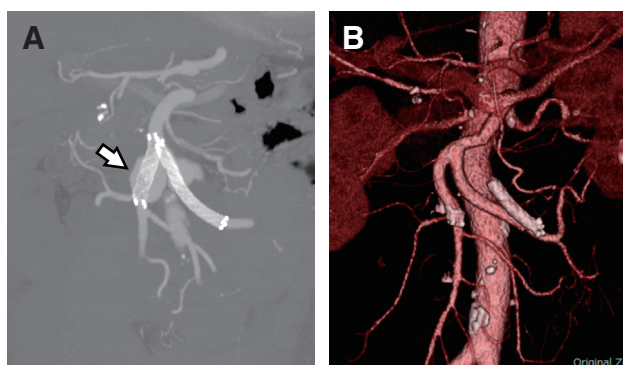


Fig. 1 Case 1: pre- and post-operative CT. (A) CT showing the superior mesenteric artery aneurysm and stents (arrow). (B) 3-dimensional CT after the infective superior mesenteric artery aneurysm resection and vascular reconstruction using an expanded polytetrafluoroethylene graft. CT: computed tomography

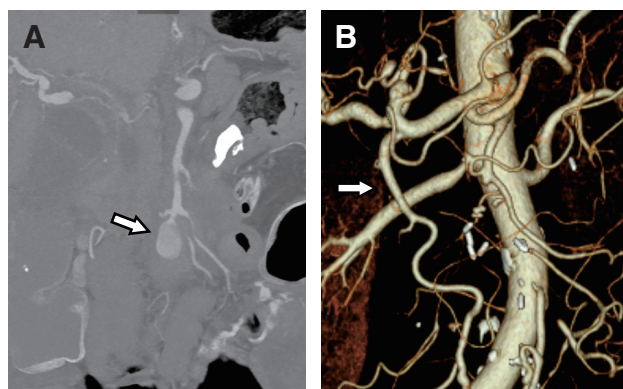


Fig. 2 Case 2: pre- and post-operative CT. (A) CT showing the SMA aneurysm (arrow) and narrowed SMA distal to the aneurysm. (B) 3-dimensional CT after the resection of the infective SMA aneurysm and vascular reconstruction. The gastroepiploic artery was anastomosed to the distal part of the SMA (arrow). CT: computed tomography; SMA: superior mesenteric artery

Case 2: A 58-year-old patient presenting with acute abdominal pain and fever was referred to our institution. Her past medical history included hypertension. CTA revealed an inflammation around the SMA, and a blood culture was taken during the admission fever workup, and the *Campylobacter fetus* was identified. Diagnosed with infective arteritis of the SMA, conservative treatment was initiated with antibiotic administration. Nonetheless, her abdominal pain persisted for two weeks, and a follow-up CTA revealed an SMA aneurysm measuring 12 mm in diameter, located approximately 2 cm distal to the origin of the middle colic artery, with narrowing of the SMA distal to the aneurysm (Fig. 2A). This prompted surgical intervention.

The procedure encompassed resection of the aneurysm and vascular reconstruction using the gastroepiploic artery as a graft¹⁾ (Fig. 2B). During the procedure, SMA was exposed over a broad area to accurately identify the vessel layers and perform appropriate anastomoses. Adhesion of the superior mesenteric vein to the SMA aneurysm was also observed.

Following the initiation of enteral nutrition, the patient experienced frequent defecation, averaging approximately 5 times per day, which was managed with loperamide. Her postoperative course was otherwise uneventful, leading to the discharge on the 15th day after surgery.

However, at the 1-month postoperative outpatient visit, the patient reported postprandial watery diarrhea. An increased dosage of loperamide and the addition of other antidiarrheal medications, such as tannate albumin and natural aluminum silicate, reduced the frequency of diarrhea from 5 to 2 times per day. However, it persisted for approximately 6 months without complete resolution. Furthermore, the patient lost her job as a cook due to hygiene concerns.

Discussion

Existing literature does not address postoperative diarrhea subsequent to the surgical intervention of infective SMA aneurysm. This may be partially attributed to the rarity of the disease, with an estimated incidence of approximately 1 case per 12000 autopsies. Infection is identified as the cause in nearly 60% of cases,²⁾ although its frequency varies across different literatures.

In the realm of gastrointestinal surgery, diarrhea has been documented as a complication following pancreatic or colon surgeries involving nerve plexus dissection around mesenteric arteries. Uncontrolled peristalsis and secretion of the denervated intestine are considered to underlie this occurrence.³⁾ In colon surgery, diarrhea improves in 6 months and does not impact the gastrointestinal quality of life.^{4,5)} However, this symptom is reported to be severe in pancreatic surgery, likely because a broader and more proximal part of the nerve plexus is dissected.⁶⁾

According to a cadaver study,⁷⁾ the neural fibers along the proximal SMA comprise 2 thick main cords localized in the loose connective tissue of the vascular sheath and finer fibers located closer to the SMA. In contrast, along the SMA distal to the retropancreatic portion, nerve fibers branch quickly, and a large number of finer fibers are relatively evenly distributed around the circumference of the vessel.

In our cases, exposing the vessel adventitia was necessary for appropriate anastomoses. During this exposure, the nerve plexus external to the adventitia might have

incurred damage. Extensive injury to this plexus, akin to that seen in pancreatic surgeries, could result in intractable diarrhea. In Case 1, intra-abdominal adhesion from prior gastrectomy and esophagectomy, coupled with ongoing infection, complicated vessel identification and exposure. Case 2 necessitated multiple anastomoses due to the aneurysm's location, potentially resulting in more significant nerve plexus injury located around the arteries.

Considering the anatomy of nerve fibers, it is imperative to avoid damaging the main cords of the nerve plexus along the proximal segment of the SMA, which could influence a substantial portion of the intestine. Therefore, vascular surgeons have to understand the aforementioned anatomy of the nerve plexus, carefully identify the main cords, and expose the adventitia with minimal incision to the vascular sheath. Although damage to the nerve fibers evenly distributed along the distal part of SMA is inevitable, the influence could be limited, given the relatively diminutive segment of the intestine innervated by these fibers.

In terms of treatment, diarrhea was reported to be manageable with both non-opioid and opioid anti-diarrheal drugs.⁶⁾ In this study, non-opioid drugs (albumin tannate, 3–6 g/day; and natural aluminum silicate, 3–6 g/day) were first administered. If these were ineffective, a dosage of 0.3 mL of opium tincture was introduced, with subsequent modification to achieve solid or soft stools 3 or fewer times a day. Notably, opioids could be discontinued within 3 years in 62% of cases, although refractory cases are not uncommon.

In our cases, loperamide, an opioid receptor agonist, exhibited partial efficacy by reducing the frequency of diarrhea but fell short of achieving complete resolution, thereby diminishing the patients' quality of life. Unfortunately, the patient in Case 1 subsequently died of uremia. In Case 2, diligent monitoring is imperative over the next 2–3 years to assess whether her symptoms ameliorate enough to consider discontinuation of loperamide. Additionally, the pursuit of more efficacious therapies capable of promptly controlling diarrhea is warranted.

Conclusion

These cases demonstrate the postoperative refractory diarrhea following the surgical intervention for infective SMA aneurysm. Given the potential deleterious impact on the quality of life, surgeons must exert utmost diligence by understanding the anatomy of the nerve plexus encircling mesenteric arteries to avoid intraoperative injury, a potential etiology of this complication. While palliative therapy

with anti-diarrheal drugs remains a viable option, expedited therapies are warranted.

Declarations

Patient consent statement

Informed consent regarding the publication of this case report is obtained from all patients.

Funding statement

None.

Ethical approval

Our institution does not require ethical approval to publish an anonymous case report.

Author contributions

Conceptualization: TN, NF

Data collection, manuscript preparation: TN

Critical review and revision: all authors

Final approval of the article: all authors

Accountability for all aspects of the work: all authors.

Disclosure statement

The authors declare no conflicts of interest for this article.

References

- 1) Fukunaga N, Shimoji A, Tamura N. In situ right gastroepiploic artery as a conduit for treating mycotic aneurysm of the superior mesenteric artery. *Eur J Vasc Endovasc Surg* 2024; **67**: 184.
- 2) Stanley JC, Wakefield TW, Graham LM, et al. Clinical importance and management of splanchnic artery aneurysms. *J Vasc Surg* 1986; **3**: 836–40.
- 3) Matsuda M, Nimura Y, Suzuki T, et al. Diarrhea after total pancreatectomy with extended dissection of the autonomic plexus. *Jpn J Gastroenterol Surg* 1982; **15**: 1765–9.
- 4) Thorsen Y, Stimec BV, Lindstrom JC, et al. Bowel motility after injury to the superior mesenteric plexus during D3 extended mesenterectomy. *J Surg Res* 2019; **239**: 115–24.
- 5) Thorsen Y, Stimec BV, Lindstrom JC, et al. Stool dynamics after extrinsic nerve injury during right colectomy with extended D3-mesenterectomy. *Scand J Gastroenterol* 2021; **56**: 770–6.
- 6) Kuroki N, Ono Y, Sato T, et al. Long-term outcome of patients with postoperative refractory diarrhea after tailored nerve plexus dissection around the major visceral arteries during pancreatoduodenectomy for pancreatic cancer. *World J Surg* 2022; **46**: 1172–82.
- 7) Reinehr MD, Vuille-Dit-Bille RN, Soll C, et al. Anatomy of the neural fibers at the superior mesenteric artery—a cadaver study. *Langenbecks Arch Surg* 2022; **407**: 2347–54.