

# Endovascular coil embolization for an anomalous splenic artery aneurysm with a splenomesenteric trunk

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## ABSTRACT

We present a case of a splenic artery (SA) aneurysm (SAA) that had arisen abnormally from the superior mesenteric artery in a 63-year-old man who underwent successful endovascular treatment. Although SAAs characterized by this anatomic abnormality are rare, in all 46 reported cases, the SAAs were located at the root of the SA and had originated abnormally from the superior mesenteric artery. This location is different from that of orthotopic SAAs, which are mostly located in the distal third of the SA. The differences in hemodynamics due to the anatomic abnormalities might play an important role in the formation of the anomalous SAAs. (*J Vasc Surg Cases Innov Tech* 2022;8:576-9.)

**Keywords:** Anomalous splenic artery; Splenic artery aneurysm; Splenomesenteric trunk; Visceral artery aneurysm

A visceral artery aneurysm (VAA) is a rare, but potentially life-threatening, disease because of the high mortality rate associated with rupture.<sup>1</sup> Although splenic artery (SA) aneurysms (SAAs) are the most common type of VAAs, SAAs with a splenomesenteric trunk are extremely rare.<sup>2,3</sup> The incidence of a splenomesenteric trunk, an anatomic anomaly in which the SA arises from the superior mesenteric artery (SMA), is quite low, occurring in only 1% of the population.<sup>4,5</sup>

We present a case of an SAA with a splenomesenteric trunk that was successfully treated endovascularly. Additionally, we reviewed the literature. The patient provided written informed consent for the report of his case details and imaging studies.

## CASE REPORT

A 63-year-old man had presented with upper abdominal pain. He had a history of hypertension and dyslipidemia. An enhanced abdominal computed tomography scan confirmed a 22-mm SAA. The SA arose from the SMA. The SAA was located at the origin of the SA and was adjacent to the back of the pancreatic head (Fig 1). Considering the risk of migration of the coil to the SMA and subsequent intestinal ischemia, the decision to initiate endovascular treatment was challenging. Informed of the risks and benefits of both open surgery and endovascular treatment, the patient decided to undergo endovascular treatment with coil embolization.

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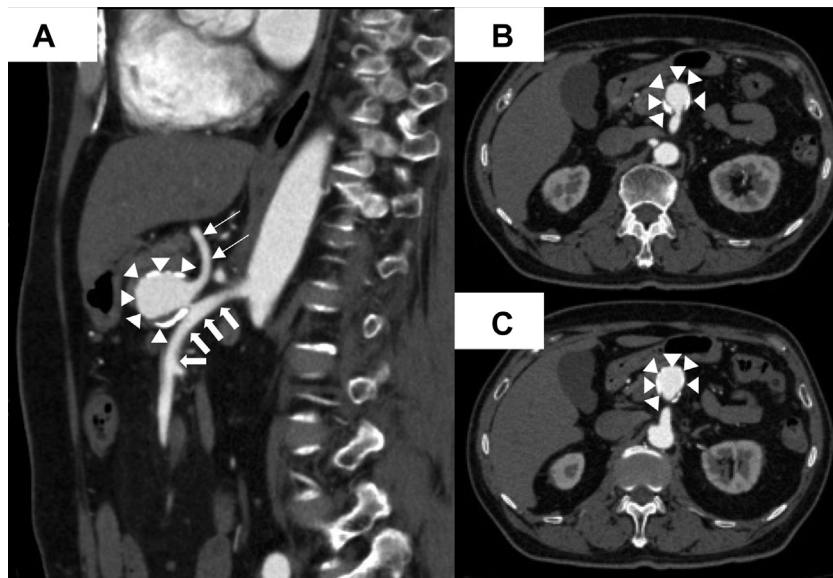
The right common femoral artery was punctured percutaneously under local anesthesia. Selective SMA angiography revealed an aneurysm located on the root of the aberrant SA that arose from the proximal SMA (Fig 2, a and b). The microcatheter was successfully threaded to the distal part of the SA, through the SAA. The SAA and the SA distal to the SAA were embolized with five coils (one 12 mm in diameter, one 9 mm in diameter, and three 6 mm in diameter; Stryker Inc, Kalamazoo, MI; Fig 2, c).

Angiography showed that the SAA was completely packed with coils, and the blood flow to the spleen was preserved through the collateral vessels. His abdominal pain had resolved the day after endovascular treatment, and he was discharged without any complications. No additional medication was provided postoperatively, including no anticoagulant or antiplatelet agents. At 3 years of follow-up after the intervention, the patient was without SAA recurrence.

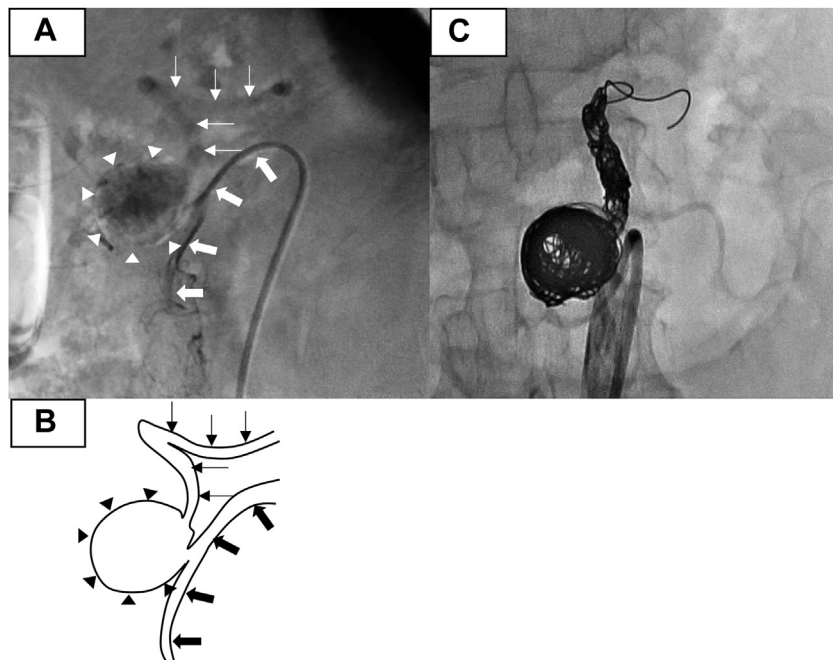
## DISCUSSION

SAAs account for 60% of all VAAs and are the third most common type of abdominal aneurysm.<sup>2,3</sup> However, the prevalence of all VAAs is 0.1% to 2%; therefore, SAAs are relatively rare compared with abdominal aortic aneurysms or iliac arterial aneurysms.<sup>1</sup> Although most SAAs will be accidentally diagnosed before the development of symptoms, once ruptured, the mortality rate has been as high as 30%.<sup>1</sup> According to the Society for Vascular Surgery guidelines, pseudoaneurysms, SAAs in patients with symptoms, and SAAs >30 mm in asymptomatic patients should be treated, although no specific recommendations has been provided for SAAs with anatomic abnormalities.<sup>6</sup>

The celiac artery is known to present with various anatomic abnormalities. Normal anatomy will be present in 85% to 89% of the population. The SMA and celiac artery arise separately from the abdominal aorta, and the celiac artery has three branches: the left gastric artery, common hepatic artery, and SA.<sup>4</sup> The abnormality in which the SA arises from the SMA, such as in the present



**Fig 1.** **a**, Sagittal view of contrast-enhanced computed tomography scan before treatment. The splenic artery (*thin arrow*) had originated from the superior mesenteric artery (SMA; *thick arrow*). The splenic artery (SA) aneurysm (SAA; *arrowheads*) was located at the root of the SA. **b,c**, Axial view of computed tomography scan showing the SAA (*arrowheads*) behind the pancreatic head.



**Fig 2.** Angiography at the site of coil embolization. **a,b**, A guiding sheath catheter was threaded to the origin of the aneurysm (*arrowhead*) through the superior mesenteric artery (SMA; *thick arrow*). *Thin arrow* indicates distal part of splenic artery (SA). **c**, The aneurysm was completely packed with coils, and the distal part of the SA was embolized.

case, is classified as a splenomesenteric trunk type. The incidence of a splenomesenteric trunk has been reported to be 1%.<sup>4,5</sup> Because of the rarity of SAAs and this anatomic abnormality, SAAs with a splenomesenteric trunk (anomalous SAAs) are extremely rare.

Since the first case reported by Chatan et al<sup>7</sup> in 1967, only 46 cases of an anomalous SAA have been reported.<sup>4,7-27</sup> A summary of the reported cases is shown in the [Table](#). The mean diameter was 32.5 mm, and 63% of the patients were women. Open resection was

**Table.** Summary of anomalous splenic artery (SA) aneurysms (SAAs; N = 46)

| Variable                         | Value        |
|----------------------------------|--------------|
| Age, years                       | 49 (22-73)   |
| Gender                           |              |
| Male                             | 17 (37.0)    |
| Female                           | 29 (63.0)    |
| Symptoms                         |              |
| Asymptomatic                     | 17 (37.0)    |
| Abdominal pain                   | 18 (39.1)    |
| Other                            | 11 (23.9)    |
| Aneurysm size, mm                | 35.5 (20-80) |
| Aneurysm location                |              |
| Origin of SA                     | 46 (100)     |
| Middle third of SA               | 0 (0)        |
| Distal third of SA               | 0 (0)        |
| Treatment                        |              |
| Surgical resection               | 21 (45.7)    |
| Surgical resection + splenectomy | 4 (8.7)      |
| Coil embolization                | 9 (19.6)     |
| Hybrid therapy                   | 2 (4.3)      |
| Stent graft placement            | 7 (15.2)     |
| Observation                      | 3 (6.5)      |
| Complications                    | 0 (0)        |

Data presented as mean (range) or number (%).

performed in 25 cases and endovascular treatment in 16 cases (nine coils and seven stent grafts), with no reported complications or deaths.<sup>4-8,11-21,23-27</sup> Since the 2000s, endovascular treatment has been applied with increasing frequency. Although the long-term outcomes of stent graft placement at the SA remain unclear, the efficacy of endovascular treatment will be sufficient if the anatomic conditions are appropriate. Thus, treating anomalous SAAs with endovascular methods can be challenging because of the risk of coil or stent graft migration to the SMA. Although the present patient had had a short aneurysmal neck, we ultimately managed to prevent coil migration by carefully using electric detachable coils. We did not use the balloon protection technique to avoid the risk of SMA injury. Given that the proximal SA of our patient was extremely short, stent graft placement in the SA was not considered. Moreover, although combined embolization of the SA distal to the SAA and stent graft placement in the SMA could also have been a treatment option, we believed that stent graft placement to the healthy SMA should be avoided if possible owing to the risk of SMA branch occlusion and the necessity for double antiplatelet treatment after the procedure.

All the reported anomalous SAA cases, as well as our case, had involved the root of the aberrant SA (Table).

In contrast, typical SAAs will most often be located in the distal third of the SA. Although the reason for this difference is unclear, it has been hypothesized that congenital alterations, such as arterial media dysplasia and sudden hemodynamic changes resulting from this anomaly, could be the primary cause of SAAs with this anatomic anomaly.<sup>18,25</sup> Thus, a risk could exist for the formation of an aneurysm at the residual neck after endovascular intervention. However, to the best of our knowledge, no studies have reported on the long-term course after treatment of this rare condition, which should be the subject for further study.

In the case of an anomalous SAA characterized by a short aneurysmal neck or an SAA close to the SMA, open resection might be more appropriate. However, when open surgical aneurysmal resection is performed, special attention should be provided to avoid pancreatic injury because anomalous SAAs will be adjacent to the back of the pancreatic head. Furthermore, surgical revascularization of the spleen must be considered in open surgery. As shown in our present case, blood flow from the collateral vessels, such as the left gastric artery, will mostly preserve splenic blood flow. Therefore, no complications associated with splenic ischemia have been reported, even in cases in which the SA was ligated or embolized without splenic revascularization.

For open surgery, a more reliable method to avoid ischemia of the spleen is to measure the blood pressure at the distal stump of the SA. We previously reported a case of an anomalous SAA surgically resected with revascularization of the SA.<sup>27</sup> In that case, the blood pressure at the distal stump was less than one half of the systemic blood pressure and requiring splenic arterial revascularization.

## CONCLUSIONS

We have presented a rare case of an anomalous SAA located at the root of the splenomesenteric trunk and treated successfully with endovascular coil embolization. This anatomic feature will influence the treatment strategy. Further studies are needed to elucidate the pathogenesis of this specific anomalous aneurysm.

## REFERENCES

1. Pitton MB, Dappa E, Jungmann F, Kloeckne R, Schotten S, Wirth GM, et al. Visceral artery aneurysms: incidence, management, and outcome analysis in a tertiary care center over one decade. *Eur Radiol* 2015;25:2004-14.
2. Obara H, Matsubara K, Inoue M, Nakatsuka S, Kuribayashi S, Kitagawa Y. Successful endovascular treatment of hemosuccus pancreaticus due to splenic artery aneurysm associated with segmental arterial mediolysis. *J Vasc Surg* 2011;54:1488-91.
3. Stanley JC, Wakefield TW, Graham LM, Whitehouse WM Jr, Zelenock GB, Lindenauer SM. Clinical importance and management of splanchnic artery aneurysms. *J Vasc Surg* 1986;3:836-40.
4. Shu C, Hu X, Luo M, Li Q, Li M, Jiang X. A review of SA aneurysms with a small subsection on aneurysms arising from a splenomesenteric trunk. *Int Angiol* 2011;30:395-407.

5. Michels NA, Siddharth P, Kornblith PL, Parke WW. Routes of collateral circulation of the gastrointestinal tract as ascertained in a dissection of 500 bodies. *Int Surg* 1968;49:8-28.
6. Chaer RA, Abularrage CJ, Coleman DM, Eslami MH, Kashyap VS, Rockman C, et al. The Society for Vascular Surgery clinical practice guidelines on the management of visceral aneurysms. *J Vasc Surg* 2020;72:3s-39s.
7. Ghatan JD, Beranbaum ER, Bowles LT, Veith FJ. Aneurysm of anomalous splenic artery. *Ann Surg* 1967;166:144-6.
8. Tanigawa N, Kariya S, Kojima H, Tokuda T, Komemushi A, Sawada S. Transcatheter coil embolization of an aneurysm of an anomalous splenic artery: usefulness of double microcatheter method. *Minim Invasive Ther Allied Technol* 2009;18:311-4.
9. Bhoil R, Tomar A, Makhaik S, Sood RC, Nayyar N. Aneurysm of an aberrant splenic artery: an extremely rare occurrence. *Vascular* 2015;23:637-8.
10. Kandoria A, Kandoria M, Ganju N, Mahajan K. Aneurysm of an anomalous splenic artery arising from splenomesenteric trunk: a rare presentation. *BMJ Case Rep* 2016;2016:bcr2015214273.
11. Manabe T, Amano T, Imoto K, Suzuki S, Kondo J, Takanaishi Y. A case of splenic artery aneurysm anomalously originating from the superior mesenteric artery. *J Jap Surg Assoc* 2000;61:1275-8.
12. Migliara B, Lipari G, Mansueto GC, Riva F, Baggio E. Managing anomalous splenic artery aneurysm: a review of the literature and report of two cases. *Ann Vasc Surg* 2005;19:546-52.
13. Tochii M, Ogino H, Sasaki H, Matsuda H, Minatoya K, Kitamura S, et al. Successful surgical treatment for aneurysm of splenic artery with anomalous origin. *Ann Thorac Cardiovasc Surg* 2005;11:346-9.
14. Sato M, Anno I, Yamaguchi M, Iida H, Orii K. Splenic artery aneurysm of the anomalous splenomesenteric trunk: successful treatment by transcatheter embolization using detachable coils. *Cardiovasc Intervent Radiol* 2006;29:432-4.
15. LaBella GD, Muck P, Kasper G, Welling R, Schlueter F, Vaughan A. Operative management of an aberrant splenic artery aneurysm: utility of the medial visceral rotation approach: a case report and review of the literature. *Vasc Endovascular Surg* 2006;40:331-3.
16. Facy O, Naouri A, Dugas B, Kadji M, Bernard P, Gabrielle F. [Aneurysm of the splenic artery arising from the superior mesenteric artery: therapeutic management]. *Ann Chir* 2006;131:401-4.
17. Illuminati G, LaMuraglia C, Nigri G, Vietri F. Surgical repair of an aberrant splenic artery aneurysm: report of a case. *Ann Vasc Surg* 2007;21:216-8.
18. Liu Q, Lu JP, Wang F, Wang L, Jin AG, Wang J, et al. Detection of anomalous splenic artery aneurysms with three-dimensional contrast-enhanced MR angiography. *Abdom Imaging* 2009;34:772-6.
19. De Cloedt L, Lavigne CM, Lardinois F, Periquet Y, Verhelst R. Aneurysm of the splenic artery which arises from the superior mesenteric artery: a case report. *Acta Chir Belg* 2010;110:332-4.
20. Wong SS, Lindsay TF, Roche-Nagle G. Anomalous splenic artery aneurysm. *Vascular* 2013;21:105-8.
21. Feo CF, Scanu AM, Fancellu A, Costantino S. Visceral aneurysm and vascular anomaly involving the splenic artery. *Dig Dis Sci* 2004;49:1378-80.
22. Mastracci TM, Cadeddu M, Colopinto RF, Cinà C. A minimally invasive approach to the treatment of aberrant splenic artery aneurysms: a report of two cases. *J Vasc Surg* 2005;41:1053-7.
23. Sakakibara K, Shindo S, Matsumoto M, Yoshida Y, Kimura M, Suzuki S, et al. Splenic artery aneurysm of the hepatosplenomesenteric trunk. *Ann Vasc Dis* 2013;6:730-3.
24. Borioni R, De Persio G, Leporace M, Di Capua C, Boggi U, Garofalo M. Endovascular treatment of multiple anomalous splenic artery aneurysms in a Jehovah witness. *G Chir* 2013;34:42-5.
25. Dong SL, Chen X, Tu ZX, Ai X, Zhang ZW, Guan Y, et al. Aneurysm of the anomalous splenic artery arising from superior mesenteric artery treated by coil embolization: a report of two cases and literature review. *Ann Vasc Surg* 2018;48:251.e5-10.
26. Kakamad FH, Hammood ZD, Salihi AM, Abdalla BY, Mohammed KS, Karim SO, et al. Aneurysm of anomalous splenic artery arising from a splenomesenteric trunk: review of the literature with a report of a new case. *Int J Surg Case Rep* 2021;80:105618.
27. Ichikawa Y, Shibutani S. A case of splenic artery aneurysm arising from superior mesenteric artery. *Jpn J Vasc Surg* 2019;28:187-92.

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