

A rare case of left colic branch aneurysm presenting with rupture and intra-abdominal hemorrhage

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ABSTRACT

An aneurysm of the inferior mesenteric artery is a rarely described clinical presentation. We have presented the case of a ruptured aneurysm originating from a branch of the inferior mesenteric artery that might represent an aneurysm of the left colic artery or the arc of Riolan. Aneurysms of this anatomic location can develop secondary to mesenteric occlusive disease, alterations in mesenteric blood flow from previous operations, or connective tissue disease. In the present case, a patient with a ruptured inferior mesenteric artery branch aneurysm had presented with intra-abdominal hemorrhage, which was successfully treated with endovascular embolization. (*J Vasc Surg Cases Innov Tech* 2021;7:447-9.)

Keywords: Aneurysm; Arc of Riolan; Coil embolization; Endovascular; Inferior mesenteric artery; Left colic; Rupture

A visceral artery aneurysm is an uncommon clinical presentation, of which colic artery aneurysms represent a small percentage. In a recent review and meta-analysis, Barrionuevo et al¹ found jejunal, ileal, and colic aneurysms constituted <1% of visceral artery aneurysms and few reported data are available describing aneurysms of the inferior mesenteric artery (IMA) or left colic arteries.² Equally infrequent is aneurysmal degeneration of the arc of Riolan, the collateral system between the superior mesenteric artery (SMA), and IMA, with only a few cases reported.³⁻⁵

In the setting of normal mesenteric blood flow, the integrity of the collateral system between the IMA and SMA such as at the arc of Riolan is often inconsequential; however, it becomes hemodynamically significant in cases of aortic or mesenteric occlusive disease.^{3,4,6} Although its presence and patency require consideration when planning aortic interventions or colon operations, these connections are infrequently the location of vascular pathology and an uncommon site of isolated intervention.³⁻⁶

We have presented a case report of a ruptured aneurysm arising from a branch of the left colic artery causing intra-abdominal hemorrhage and treated successfully

with embolization. The patient provided written informed consent for the report of her case. We believe our report will contribute to the limited data on this topic, and, unlike most previous reports, our patient had no significant aortic or mesenteric atherosclerosis and no relevant previous surgeries.³⁻⁵

CASE REPORT

A 45-year-old woman, a former smoker with α_1 -antitrypsin deficiency (AAT) and chronic obstructive pulmonary disease, who was listed for a lung transplant, had presented to the emergency department with a several-day history of abdominal pain and shortness of breath. Her surgical history included anterior and posterior fusion of the fourth and fifth lumbar vertebrae and right hemilaminectomy and discectomy of the second and third lumbar vertebrae via a posterior approach.

On examination, she was afebrile with normal blood pressure but a heart rate of 150 bpm. The pertinent laboratory data included a white blood cell count of $14.3 \times 10^3/\mu\text{L}$, platelet count of $482 \times 10^3/\mu\text{L}$, and an international normalized ratio of 1.0. Her hemoglobin was initially 10.7 g/dL, which was decreased compared with a recent outpatient value of 12.1 g/dL. After fluid resuscitation, her hemoglobin had decreased to 8.7 g/dL. The physical examination revealed diffuse abdominal tenderness and increasing distention.

Contrast-enhanced computed tomography of the chest, abdomen, and pelvis demonstrated a ruptured 9-mm focal aneurysm that appeared to arise from a branch of the IMA and was associated with a 12-cm mesenteric hematoma. No significant atherosclerotic disease was present in the aorta or mesenteric vessels (Fig 1).

The patient was taken for emergent mesenteric angiography with embolization. Arterial access was obtained via ultrasound-guided puncture of the right common femoral artery, and a 6F sheath was placed. The SMA was selectively cannulated, and the angiographic findings appeared normal. The IMA was then selected, and angiography confirmed a focal aneurysm of the first branch of the left colic artery or the arc

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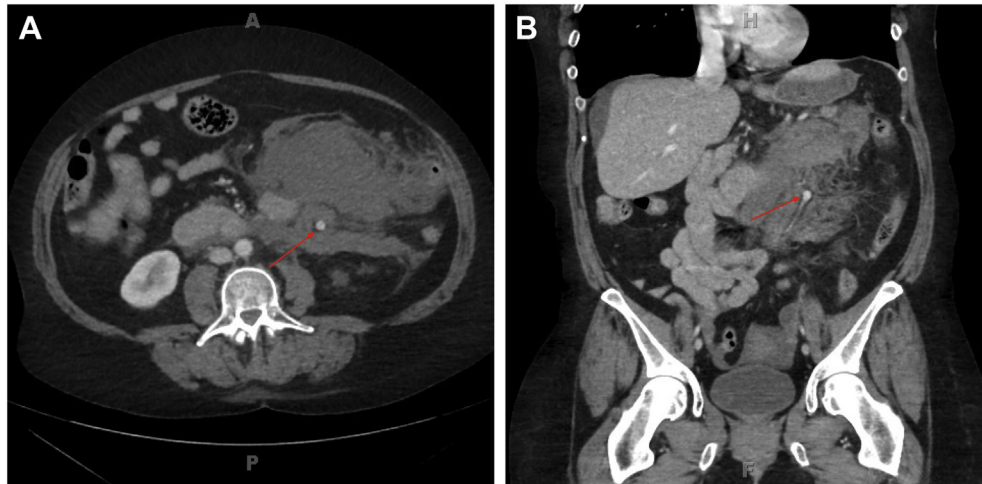


Fig 1. Contrast-enhanced computed tomography scan on presentation showing 9-mm aneurysm (red arrow) from an inferior mesenteric artery (IMA) branch vessel and surrounding intra-abdominal hematoma on axial (**A**) and coronal (**B**) series.

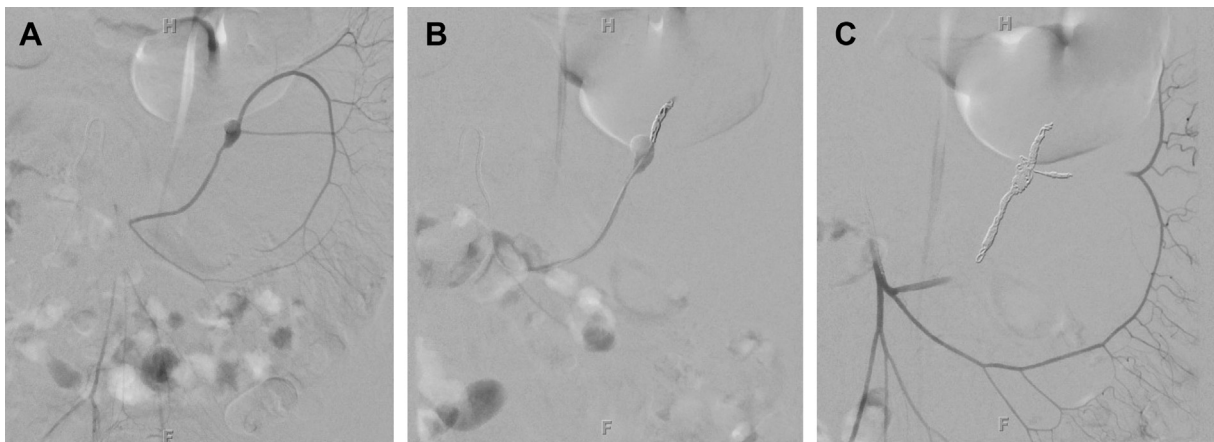


Fig 2. **A,** Selective inferior mesenteric artery (IMA) angiogram demonstrating focal aneurysm of a branch of the IMA. **B,** Embolization of an out-flow vessel. **C,** Successful completion angiogram after embolization.

of Riolan. An intact marginal artery of Drummond was noted. A Progreat microcatheter (Terumo Medical, Tokyo, Japan) was advanced into the arc, and sequential embolization was performed of the two out-flow vessels, aneurysm, and in-flow vessel using Ruby coils (Penumbra, Alameda, Calif). Completion IMA angiography demonstrated successful embolization (Fig 2).

The patient did well postoperatively and had no bowel ischemia complications. This lesion was believed to be low risk owing to the location, the intact marginal artery of Drummond, and the lack of underlying stenosis of the mesenteric vessels. To exclude other vasculopathy, cervical imaging was obtained, which demonstrated a subtle corrugated appearance of the bilateral internal carotid arteries. This finding led to a presumed diagnosis of fibromuscular dysplasia (FMD), which was supported by the small amount of data available regarding concomitant FMD and AAT deficiency.⁷

The patient recovered well and was discharged in stable condition. At the 2-month follow-up, multiphasic computed tomography demonstrated successful embolization and a reduced size of the intra-abdominal hematoma to 7.8 cm.

DISCUSSION

It is well accepted that the arc of Riolan and other direct connections between the IMA and SMA play a critical role in the collateral circulation in the setting of an abnormal mesenteric arterial supply, including cases of occlusive disease or after either operative ligation or endovascular exclusion of the IMA.^{3,4} In these cases, these collateral networks enlarge to compensate for the arterial occlusions. Xie et al⁸ demonstrated a significant increase in the diameter of the arc in patients with SMA or IMA occlusive disease compared with controls.

It is this ectasia of the vessel and hyperdynamic flow that are among the proposed mechanisms of visceral artery aneurysm formation and the causes of aneurysms elsewhere in the body, including atherosclerosis, trauma, and connective tissue disorders.³ Two of the three previously reported case reports describing arc of Riolan aneurysms involved patients with an apparent cause for the hyperdynamic flow: one with atherosclerotic disease of the celiac trunk and SMA and one after aortic endograft with IMA coverage.^{3,4} The third case shared features with our case, in that the patient did not have atherosclerotic disease and not undergone a previous aortic intervention.⁵

Left colic artery aneurysms are similarly infrequent, with few cases reported, and little has been elucidated on their etiology and natural history.² Of the available data on both ruptured arc of Riolan and left colic aneurysms, most had undergone successful endovascular intervention with embolization.²⁻⁵

In planning for embolization, consideration must be given to the potential risk of bowel ischemia, and it can be assumed that the risk will depend on the underlying disease process. Specifically, for patients with mesenteric stenosis or disruption of the normal vasculature from previous surgery, intestinal perfusion could be largely dependent on increased flow through the branches of the IMA, and such patients likely have an increased risk of ischemia after embolization.¹ In contrast, for patients without mesenteric disease, such as our patient, the assumed risk of bowel ischemia should be low, especially in the presence of other collateral networks, such as an intact marginal artery of Drummond.

Our patient's vasculopathy evaluation demonstrated features consistent with FMD. FMD is a noninflammatory, nonatherosclerotic arteriopathy resulting in stenosis and aneurysms, most commonly affecting the carotid and renal arteries. However, rare cases of mesenteric involvement have been described.^{9,10} Additionally, our patient's history of AAT deficiency warranted consideration, because deficiency of the elastase inhibitor has been implicated as a possible etiology of aneurysmal degeneration via proteolysis of arterial proteins and has been rarely reported as a cause of visceral artery aneurysms.^{11,12} A limited volume

of data are available on the occurrence of concomitant FMD and AAT; however, some association between the two disorders has been proposed.^{7,12}

CONCLUSIONS

In the present report, we have described a case of a ruptured left colic branch aneurysm treated with endovascular embolization, adding to the small body of existing data on the topic. In contrast to some previous reports, this occurred in the absence of mesenteric atherosclerotic disease or previous aortic intervention but, rather, in a patient with a history of AAT deficiency and newly diagnosed FMD.

REFERENCES

1. Barrionuevo P, Malas MB, Nejm B, Haddad A, Morrow A, Ponce O, et al. A systematic review and meta-analysis of the management of visceral artery aneurysms. *J Vasc Surg* 2019;70:1694-9.
2. Kota AA, Wang K, Leckie K, King J, Majjub J, Motaganahalli RL. Left colic artery aneurysm. *J Vasc Surg* 2020;72:1457-8.
3. Kumaresh A, Rajoo R, Babu SR, Ilanchezhian S. A rare case of aneurysm of arc of Riolan artery and gastroduodenal artery. *J Clin Imaging Sci* 2014;4:66.
4. Liu X, Zhang J, Chen H, Zhang L, Wang H, Ji M, et al. Riolan arch pseudoaneurysm hemorrhage after endovascular covered stent-graft treatment of an abdominal aortic aneurysm: a case report. *Medicine (Baltimore)* 2019;98:e17789.
5. Chen YT, Hsu MY, Lee YH, Tseng JH. Embolization of an arc of Riolan artery aneurysm. *J Vasc Interv Radiol* 2020;31:1320.
6. van Gulik TM, Schoots I. Anastomosis of Riolan revisited: the meandering mesenteric artery. *Arch Surg* 2005;140:1225-9.
7. Schievink WI, Björnsson J, Parisi JE, Prakash UB. Arterial fibromuscular dysplasia associated with severe alpha 1-antitrypsin deficiency. *Mayo Clin Proc* 1994;69:1040-3.
8. Xie Y, Jin C, Zhang S, Wang X, Jiang Y. CT features and common causes of arc of Riolan expansion: an analysis with 64-detector-row computed tomographic angiography. *Int J Clin Exp Med* 2015;8:3193-201.
9. Kimura K, Ohtake H, Kato H, Yashiki N, Tomita S, Watanabe G. Multivisceral fibromuscular dysplasia: an unusual case of renal and superior mesenteric involvement. *Ann Vasc Dis* 2010;3:152-6.
10. St Jean P, Hart B, Webster M, Steed D, Adamson J, Powell J, et al. Alpha-1-antitrypsin deficiency in aneurysmal disease. *Hum Hered* 1996;46:92-7.
11. Mitchell MB, McAnena OJ, Rutherford RB. Ruptured mesenteric artery aneurysm in a patient with alpha 1-antitrypsin deficiency: etiologic implications. *J Vasc Surg* 1993;17:420-4.
12. Bofinger A, Hawley C, Fisher P, Daunt N, Stowasser M, Gordon R. Alpha-1-antitrypsin phenotypes in patients with renal arterial fibromuscular dysplasia. *J Hum Hypertens* 2000;14:91-4.

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