

Case report of suspected Takayasu arteritis manifesting as chronic mesenteric ischaemia: a rare cause for a typical clinical picture

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ABSTRACT

INTRODUCTION: Chronic intestinal ischemia accounts for approximately 1:1,000,000 admissions in some epidemiological studies. This condition usually affects individuals with known atherosclerotic lesion in other territories. Although typically found in female patients in their seventh decade of life with established arterial disease, chronic intestinal ischemia may manifest itself in younger patients. In the latter case, unusual causes, such as vasculitis, must be included in the differential diagnosis.

CASE REPORT: We report the case of a 31-year-old man with a known history of intermittent left and right upper limb claudication and smoking. He complained of postprandial epigastric pain in the previous year, with involuntary weight loss (6kg in the previous 6 months) and fear of eating. An angio CT scan was performed, which showed occlusion of the celiac trunk and superior mesenteric artery at its origin. The diagnosis of chronic intestinal ischemia was formulated, and the patient was submitted to an antegrade bypass from the supraceliac aorta to the superior mesenteric artery and common hepatic artery, with a bifurcated Dacron graft.

DISCUSSION: Given the inflammatory nature of the collected fragment of aorta, the onset of intestinal ischemia at a young age and the remaining peripheral arterial manifestations, a presumptive diagnosis of Takayasu's arteritis was formulated. This vasculitis is typically found in patients in their third decade and is associated with diffuse arterial thickening. Mesenteric manifestations in this condition can occur up to 30%.

Keywords: intestinal ischemia; mesenteric ischemia; vasculitis; Takayasu arteritis.

INTRODUCTION

Chronic intestinal ischemia is a clinical entity defined by a hemodynamic failure in the mesenteric circulation, predominantly in the postprandial period. It is responsible for approximately 1:1000000 hospital admissions in some epidemiologic series.^[1] This condition commonly affects individuals with known atherosclerotic lesion in other territories. Symptoms typically arise when 2 of the main mesenteric vessels are affected. Although typically found in female patients in their seventh decade with established

arterial disease, chronic intestinal ischemia may manifest in younger patients with no or few cardiovascular risk factors. In the latter, uncommon causes, such as vasculitis, are to be included in the patient workup. This case illustrates the need for an extensive differential diagnosis in atypical patients.

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CASE REPORT

We report the case of a 31-year-old male, emigrated from Bulgaria since 2011. This patient is an active smoker (over 15 pack-years), with no other known cardiovascular risk factors. He has a known history of dyspepsia and recurring epigastric pain. An esophagogastroduodenoscopy (EGD) was performed, which revealed the presence of *H.pylori*, which was treated with an appropriate antibiotic course. Since 2017, the patient complained of left lower limb and right upper limb intermittent claudication. A lower limb CT scan was performed, showing right popliteal artery occlusion and diffuse tibioperoneal arterial disease. Over the last consultations, he complained of severe postprandial upper abdominal pain in the previous year, with concomitant unvoluntary weight loss (6kg in the previous 6 months) and fear of eating. Physical examination was performed, and the abdomen was mildly tender when palpating the epigastrium, with no signs of peritonitis. Right radial pulse was diminished and left popliteal and distal pulses were abolished. Laboratory workup was unremarkable, except for an elevated SR (31mm; normal range < 10). A CT angiography was performed, showing occlusion of the celiac trunk and superior mesenteric artery at their origin (figure 1).

Figure 1. preoperative CT angiogram.

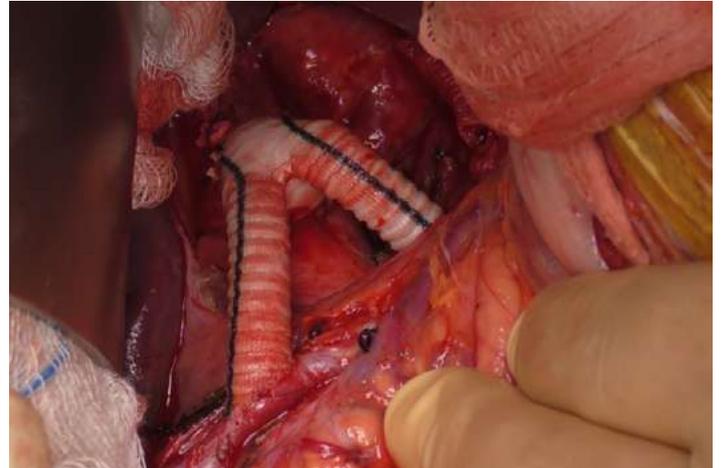


Complete ostial occlusion of the celiac trunk and superior mesenteric artery is evidenced (white arrows).

There was no significant disease in the supraceliac aorta, with no wall thickening or inflammatory signs in other segments. Serologic markers of autoimmunity or thrombophilia were all negative. The patient was admitted to the vascular surgery ward for etiological investigation and surgical repair of the mesenteric vessels. Given the age and atypical clinical background, the case was discussed with the rheumatology team. A supra-aortic arterial ultrasound was performed, showing thickening of the right temporal artery, as well as mild thickening of common carotid arteries bilaterally. Since the patient had no major comorbidities, with a seemingly

healthy supraceliac aorta, the surgical team decided to perform an antegrade bypass from the supraceliac aorta to the superior mesenteric artery and common hepatic artery, with implantation of a bifurcated Dacron graft prosthesis (Figure 2).

Figure 2. Intraoperative aspect of the revascularization.



Antegrade bypass from the supraceliac aorta to the superior mesenteric artery and common hepatic artery *in situ*.

The surgical procedure was performed under general anesthesia with no major perioperative complications. A small fragment of the aorta and periaortic lymph node were collected for analysis. Anatomico-pathological results of the aortic fragment showed the presence of fibrous and muscular tissue with a mild, mixed inflammatory infiltrate. The lymph node appeared to have reactive inflammatory cells. The post-operative period was uneventful (figure 3), and the patient was discharged home 6 days after surgery. During follow-up, the patient reported no further complaints, with no postprandial pain.

Figure 3. Postoperative CT reconstruction.



A patent antegrade bypass *in situ* is shown (white arrow).

DISCUSSION

This patient illustrates a typical presentation of chronic intestinal ischemia. Indeed, he complained of severe postprandial upper abdominal pain with involuntary weight loss and fear of eating. These symptoms reflect an inappropriate supply in the mesenteric circulation in a postprandial setting, underlying severe mesenteric disease as shown in the CT scan. In this case, the diagnosis could have been accurately confirmed by duplex ultrasound. Although this exam is highly operator-dependent, it is the first-line screening exam to dynamically assess flow in mesenteric vessels.^[2] Nevertheless, apart from the history of intermittent claudication and smoking habits, this patient had no other relevant cardiovascular risk factors. Furthermore, with these symptoms at a young age, typical atherosclerotic causes do not seem to be a major factor to this condition. Given the inflammatory nature of the collected fragment of aorta, the onset of intestinal ischemia at a young age and the remaining peripheral arterial manifestations, a presumptive diagnosis of Takayasu's arteritis was formulated. This vasculitis is typically found in patients in their third decade and is associated with diffuse arterial thickening. Mesenteric manifestations in this condition can occur up to 30%. When reviewing the ACR clinical criteria for Takayasu arteritis, this patient fulfills three criteria (age, limb claudication and diminished brachial pulse).^[3] To achieve a definitive diagnosis, imaging exams such as ultrasound, MRI or PET scan are of great importance to identify arterial inflammatory foci and disease extension and activity.^[4] Regarding treatment, immunosuppressive agents such as systemic corticoids, methotrexate or cyclophosphamide are the mainstay. Surgical treatment in these patients is indicated in extensive and occlusive arterial disease, is associated with better long-term results, namely higher patency rates.^[2,5] Overall, endovascular treatment of patients with chronic intestinal ischemia is associated with decreased morbidity, length of stay, and convalescent time.^[5-7] Nevertheless, in patients with Takayasu arteritis, open surgery is usually associated with lower restenosis rates compared with endovascular treatment, with similar mortality and complication rates.^[8-9]

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REFERENCES:

1. Schermerhorn ML, Giles KA, Hamdan AD, Wyers MC, Pomposelli FB. Mesenteric revascularization: management and outcomes in the United States, 1988-2006. *J Vasc Surg* 2009;50(2):341-8.
2. Björck M, Koelemay M, Acosta S, Bastos Goncalves F, Kölbl T, Kolkman JJ, et al. Management of the Diseases of Mesenteric Arteries and Veins. *Eur J Vasc Endovasc Surg* 2017;53(4):460-510.
3. Arend WP, Michel BA, Bloch DA, Hunder GG, Calabrese LH, Edworthy SM, et al. The American College of Rheumatology 1990 criteria for the classification of takayasu arteritis. *Arthritis Rheum* 1990;33(8):1129-34.
4. Dejaco C, Ramiro S, Duftner C, Besson FL, Bley TA, Blockmans D, et al. EULAR recommendations for the use of imaging in large vessel vasculitis in clinical practice. *Ann Rheum Dis*. 2018;77(5):636.
5. Huber TS, Björck M, Chandra A, Clouse WD, Dalsing MC, Oderich GS, et al. Chronic mesenteric ischemia: Clinical practice guidelines from the Society for Vascular Surgery. *J Vasc Surg*. 2021;73(1):87S-115S.
6. Oderich GS, Bower TC, Sullivan TM, Bjarnason H, Cha S, Glociczki P. Open versus endovascular revascularization for chronic mesenteric ischemia: Risk-stratified outcomes. *J Vasc Surg*. 2009;49(6):1472-9.
7. van Petersen AS, Kolkman JJ, Beuk RJ, Huisman AB, Doelman CJA, Geelkerken RH. Open or percutaneous revascularization for chronic splanchnic syndrome. *J Vasc Surg*. 2010;51(5):1309-16.
8. Rits Y, Oderich GS, Bower TC, Miller DV, Cooper L, Ricotta JJ, et al. Interventions for mesenteric vasculitis. *J Vasc Surg*. 2010;51(2):392-400.
9. Jung JH, Lee YH, Song GG, Jeong HS, Kim J-H, Choi SJ. Endovascular Versus Open Surgical Intervention in Patients with Takayasu's Arteritis: A Meta-analysis. *Eur J Vasc Endovasc Surg*. 2018;55(6):888-99.