Case Report

Spontaneous isolated celiac and splenic artery dissection with splenic infarction

Shaun Pateman Aciu, BS<sup>a</sup>,*, Jameson Petrochko, MD<sup>b</sup>, Noy Bassik, MDPhD<sup>c</sup>, Jay Fisher, MD<sup>d</sup>

<sup>a</sup>Temple/St. Luke’s School of Medicine, 800 Ostrum St., Bethlehem, PA 18015, USA
<sup>b</sup>Department of Surgery, St. Luke’s University Health Network, Bethlehem, PA, USA
<sup>c</sup>Department of Interventional Radiology, St. Luke’s University Health Network, Bethlehem, PA, USA
<sup>d</sup>Department of Vascular Surgery, St. Luke’s University Health Network, Bethlehem, PA, USA

Abstract

Spontaneous isolated celiac artery dissection is considered an uncommon clinical condition. Rarer still is spontaneous isolated celiac and splenic artery dissection, with a total of 42 reported cases. There is no known definitive cause of visceral artery dissections, but risk factors include male sex, age in 5th or 6th decade, hypertension, and connective tissue disorders. The presentation varies, diagnosis is principally radiographic, and the mainstay of treatment is anticoagulation or antiplatelet therapy. Splenic infarction is a common finding with splenic artery dissection, although the strength of this association has not previously been reported. Herein we present a case of spontaneous isolated celiac and splenic artery dissection with splenic infarction that was successfully managed with blood pressure control and antiplatelet therapy. We review previous literature, principles of diagnosis and management, and incidence and outcomes of splenic infarction as it related to splenic artery dissection.

© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington

This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

The main visceral arteries are the celiac, superior mesenteric, and inferior mesenteric arteries. Dissection of these arteries most commonly originates from the aorta and extends into these arteries. When the dissection originates within visceral artery itself, not involving the aorta, it is called a spontaneous isolated dissection of the artery involved. Collectively, these processes are called spontaneous isolated visceral artery dissections. This is a relatively rare occurrence. Involvement of the SMA is most common, followed by the celiac artery [19]. The dissection can spread along the involved vessel’s wall and involve its branches, including the splenic artery.

Spontaneous isolated celiac artery dissection is considered an uncommon clinical condition. Morgan et al. reported roughly 200 documented cases as of 2018 [11]. Rarer still is spontaneous isolated celiac and splenic artery dissection.
Zenda et al. reported 23 “appropriately documented” cases of splenic artery dissection as of 2017, not including their case [25]. Herein we present a case of spontaneous isolated celiac and splenic artery dissection with splenic infarction that was successfully managed with blood pressure control and ant-iplatelet therapy. We review previous literature, principles of diagnosis and management, and incidence and outcomes of splenic infarction as it related to splenic artery dissection.

Case report

A 57-year-old man presented to the emergency department (ED) with 6 hours of acute-onset post-prandial abdomi- nal pain. His hypertension was normally well-controlled on losartan-hydrochlorothiazide and he had not missed any doses recently. His initial blood pressure in the ED was 197/93 mmHg with sinus bradycardia at 57 BPM via EKG and otherwise normal vital signs. Medical history included left or- chectomy 10 years prior for testicular cancer. He had no history of heart disease, smoking, alcohol consumption or other cardiac risk factors. Laboratory investigations revealed mild leukocytosis at 12,610 WBCs/µl with 84% neutrophils. Complete metabolic panel, Troponin I, Lactic Acid, and Lipase were all within normal limits.

Computer tomography angiogram (CTA) of the chest/abdomen/pelvis was performed as there was a sus- picion of aortic dissection. The study revealed dissection in the celiac trunk with a small amount of contrast flow noted within a false lumen. The dissection extended throughout most of the length of the splenic artery with subsequent segmental areas of splenic infarction. The dissection flap did not extend into the hepatic or gastric artery branches. The median arcuate ligament angle appeared within normal limits (Figs. 1.1, 1.2, 1.3, 1.4, 1.5). A 4.5 cm ascending aortic aneurysm was noted as well.

Approximately 3 months prior, the patient underwent a CT chest/abdomen/pelvis with contrast for screening of testicular cancer metastases. The CT at this time was performed in the tissue phase. Retrospective analysis of the celiac and splenic arteries showed no evidence of dissection or other abnormal anatomic variants (Fig. 2). There was a 4.5cm ascending aortic aneurysm, which was noted as stable in the present CT.

Blood pressure control was initiated in the ED with labetalol and hydralazine. Pain was also controlled with acetaminophen, oxycodone, and hydromorphone. The patient was admitted to the hospitalist service with vascular surgery consultation. The patient was managed non-operatively with blood pressure control, dual anti-platelet therapy (aspirin and clopidogrel), and serial abdominal exams. The patient’s pain improved and he was discharged home in good condition on hospital day two with outpatient follow up.

The patient has remained symptom-free in the interval pe- riod between presentation and follow up imaging. Follow up CT angiogram at 6 months demonstrated marked interval im- provement of the dissection with only a very small focal dis- section of the distal celiac artery that did not extend into the celiac branch vessel (Fig. 3). The splenic artery was grossly patent without abnormality. The spleen demonstrated normal

Fig. 1.1 – Coronal CTA demonstrating dissecting celiac artery with thrombosed false lumen.

Fig. 1.2 – Coronal CTA demonstrating axial view of dissecting splenic artery with thrombosed false lumen. The intact common hepatic artery is also visualized.
heterogeneity without evidence of infarction. Blood pressure at follow up was 100/70. The vascular team recommended to continue non-operative management with blood pressure control indefinitely and aspirin therapy only.

Discussion

It is widely believed that reports of all spontaneous isolated visceral artery dissections will continue to increase over time.
as diagnostic imaging modalities improve and become more prevalent [6,25]. Indeed, our search found 18 additional cases of splenic artery dissection, bringing the number of reported cases to 42, including our case [3-6,8,9,24]. Males in 5th or 6th decade of life are at highest risk. Other risk factors include hypertension, sleep apnea, smoking, trauma, and connective tissue disease. Spontaneous isolated visceral artery dissections are not believed to be related to atherosclerosis [6,19].

The etiology of all spontaneous isolated visceral artery dissections remains unclear. It has been suggested that compression of the celiac artery by the median arcuate ligament may play a role in spontaneous isolated celiac artery dissection [6]. Additionally, as Park et al. demonstrated, variations in flow dynamics based on vascular anatomy can have profound effect on risk of spontaneous isolated superior mesenteric artery (SMA) dissection, a similar condition [17]. We wonder if anatomic variations also predispose the celiac artery to spontaneous dissection in some patients. Furthermore, we question what causes the dissection to spread into the splenic artery in some patients but not others. We hypothesize that anatomic variations in the characteristically tortuous splenic artery could predispose extension of dissection into the vessel in some patients [18]. More detailed study of splenic artery anatomy in patients with splenic artery dissection is warranted. Still, most cases of spontaneous isolated visceral artery dissection remain idiopathic.

The clinical presentation of splenic artery dissection is variable, ranging from asymptomatic to life threatening [2,3,11,16]. If symptomatic, patients often present with nonspecific abdominal, back, flank, or chest pain, and nausea/vomiting [3,5,9,11,12,25]. It is occasionally associated with physical activity, such as jogging and golfing, but this appears to be infrequent [12,13,24]. As was seen with our patient, presentation in hypertensive urgency has been reported [3,12,14]. This presentation is not universal, as patients have been reported as normotensive [10] or mild to moderately hypertensive [2,22,25] upon presentation.

The diagnosis of spontaneous isolated visceral artery dissection is radiographic, typically with CT with contrast or CTA. The diagnosis is made when an intimal flap with enhancing false lumen or a non-enhancing crescent-shaped area along the wall of an artery is seen. The former indicating a double-
Barreled dissection and the latter indicating a thrombosed false lumen [17,20]. Multiple classification systems have been proposed for spontaneous isolated SMA dissection, but not for spontaneous isolated celiac artery dissection [7,20]. Acute splenic infarction typically appears as a peripheral, wedge-shaped, non-enhancing defect [21]. In the reported cases of splenic artery dissection, the largest reported splenic infarct was estimated at 60% [13].

There is not a consensus on how best to manage patients with splenic artery dissection. Its treatment is like that of all visceral artery dissections. Most patients are managed medically, consisting of either anti-platelet, anti-coagulation, both, or with observation alone. Anti-hypertensive and pain control regimens are used when indicated [4,6,8,9,12,13,20,22–25]. In a review of 77 patients with either spontaneous isolated SMA or celiac artery dissection, Morgan found that asymptomatic patients tended to be treated with anti-platelet therapy alone while symptomatic patients were treated with anticoagulation [11]. Surgical or endovascular intervention are reserved for patients with concomitant splenic artery hematoma or aneurysm [3,10,15]. Splenectomy was performed in two patients who had splenic infarction, one of which had a history of Factor V Leiden [13,16].

It is not yet clear what follow-up imaging guidelines should be for spontaneous isolated visceral artery dissection. Hosaka et al. suggest follow up imaging should be obtained at 1 month and every 3 to 12 months thereafter for “a long period,” citing a concern for development of aneurysm formation in the months to years after initial dissection [6]. However, Park et al. found CT follow-up of patients with spontaneous isolated SMA or celiac artery dissection was only useful within the first week of presentation. In a study of 33 patients, they found no significant radiologic changes occurring from the first week to six months after presentation and patient symptoms did not seem to correlate with imaging findings [17]. Given the lack of sufficient data or guidelines, it is up to the providers’ discretion as to when, how often, and for how long to obtain follow-up imaging in patients with spontaneous isolated visceral artery dissection.

As the ability to diagnose and treat splenic artery dissection has improved, so too has its outcomes. The diagnosis was initially made post-mortem [2]. Nowadays, patients tend to

**Fig. 3** – CTA abdomen 6 months after dissection demonstrating stable small dissection at distal celiac artery with patent celiac artery branches.
have favorable outcomes, with resolution of symptoms and minimal reported further complications. Except for the two aforementioned cases of splenectomy, patients appear to recover well from splenic infarct without long term sequelae [3,4,6,8–10,12,14,15,20,23–25]. Follow up imaging has demonstrated stable dissection and splenic scarring as far out as 2 years from presentation [24].

It is unclear how common splenic infarction is in the setting of splenic artery dissection. To estimate the frequency of concomitant splenic infarction, we reviewed the 42 reported cases of spontaneous isolated celiac and splenic artery dissection to assess the imaging findings of the spleen. The 7 patients reported from Gao et al. were not included in the analysis because it was not specified which patients had splenic infarction [5], 13 of the cases were diagnosed on autopsy with no imaging obtained [2]. Of the remaining 22 cases, 14 reported splenic infarctions [3,4,6,8,12,13,16,20,23,24], 4 reported no infarction [6,10,14,25], and the remaining 4 did not mention imaging of the spleen [9,15,20,22]. Excluding the 13 cases diagnosed on autopsy and assuming the 4 patients with no mention of splenic imaging did not have splenic infarction, we calculate that 14/22 (64%) of patients with spontaneous isolated celiac artery and splenic artery dissection have concomitant splenic infarction. It should be noted that involvement of the splenic artery is not necessary for splenic infarction [1]. Gao et al. reported splenic infarction in 18.92% of patients presenting with celiac artery dissection [5], not all of which involved the splenic artery.

While our patient does have a history of hypertension, review of his primary care records revealed well controlled in-office blood pressures in the 120s/70s. The patient reported near 100% compliance with his home medication regiment and did not report miss any dosages in the days leading up to his presentation. This is at odds with the patient’s presentation in hypertensive urgency. The aortic aneurysm found on CT is not thought to have played a role in the patient’s current presentation, given that it was stable over time and was limited to the ascending aorta.

Serendipitously, our patient obtained a CT scan with contrast for a 10-year testicular cancer screening 3 months prior to his current presentation which revealed no evidence of arterial dissection. Our case exemplifies the possibility of an acute, unpredictable presentation. This is in contrast with reports of patients presenting asymptotically [11,19,20,25]. Remarkably, one patient with spontaneous isolated celiac artery dissection without splenic artery involvement presented asymptotically and was stable for 101 months [6].

Conclusion

Spontaneous Isolated Celiac and Splenic Artery Dissection remains a rare clinical entity. However, as more patients are imaged and image quality improves, this diagnosis will become more frequent. Providers should be aware of this diagnosis, its variable presentation, and its management, which is often conservative.

_____

Patient consent

Written informed consent was obtained from the patient regarding publication of their case. This documentation is kept on record.

REFERENCES

[1] Abougroun Ashraf, Natarajan Arjun, Daoud Hussein, Khalaf Habeeb. Spontaneous celiac artery dissection presenting with splenic infarction: a case report. Gastroenterol Res 2018;11(5):379–82. doi:10.14740/gr1065w.
[2] Desinan Lorenzo, Scott Cathryn A, Piai Iginio, Mazzolo Gionata. Sudden death due to spontaneous rupture in splenic artery atypical dissection with features of vasculitis: case report and review of the literature. Forensic Sci Int 2010;200(1–3). doi:10.1016/j.forsciint.2010.02.035.
[3] Ebaugh James L, Chew David K. Coil embolization of an acutely expanding spontaneous splenic artery dissection. J Vasc Surg Cases Innov Tech 2018;4(2):144–6. doi:10.1016/j.jvscit.2018.01.010.
[4] Emori Kazumasa, Takeuchi Nobuhiro, Soneda Junichi. A case of isolated celiac artery dissection accompanied by splenic infarction detected by ultrasonography in the emergency department. Case Rep Cardiol 2016;2016:1–5. doi:10.1155/2016/8608496.
[5] Gao Xiujuan, Xie Weiping, Shang Dan, Liu Jianyong, Jin Bi, Xu Ying. Clinical features and management strategy of symptomatic spontaneous isolated celiac artery dissection. Vascular 2021;170853812098629. doi:10.1177/170853812098629.
[6] Hosaka Akihiro, Nemoto Masaru, Miyata Tetsuro. Outcomes of conservative management of spontaneous celiac artery dissection. J Vasc Surg 2017;65(3). doi:10.1016/j.jvs.2016.09.058.
[7] Jia Zhongzhi, Tu Jianfei, Jiang Guomin. The classification and management strategy of spontaneous isolated superior mesenteric artery dissection. Korean Circ J 2017;47(4):425–31. doi:10.4070/kcj.2016.0237.
[8] Lee Seo I, Kim Jin J, Yang Hyuk J, Lee Keun, et al. Spontaneous dissection of celiac trunk with concurrent splenic artery dissection. Clin Exp Emerg Med 2015;2(4):256–9. doi:10.15441/ceem.15.030.
[9] Melnychuk Eric, Strony Robert. Spontaneous isolated visceral artery dissection in a middle aged male. Case Rep Emerg Med 2017;2017:1–3. doi:10.1155/2017/3704348.
[10] Merrell Steven W, Gloviczki Peter. Splenic artery dissection: a case report and review of the literature. J Vasc Surg 1992;15(1):221–5. doi:10.1016/0741-5214(92)70032-g.
[11] Morgan Courtney E, Mansukhani Neel A, Eskandari Mark K, Rodriguez Heron E. Ten-year review of isolated spontaneous mesenteric arterial dissections. J Vasc Surg 2018;67(4):1134–42. doi:10.1016/j.jvs.2017.08.071.
[12] Moussa Tania, Nawfal Georges, Assi Tarek, Rassay Elie E, Massoud Elie, Daou Iskandar. Isolated celiac artery dissection: a case report and review of the literature. Case Rep Vasc Med 2015;2015:1–3. doi:10.1155/2015/194079.
[13] Muñoz-García Ángela, Abarca-Costalago Manuel, Hierro-Martín Isabel, Valdivieso-Felices Pedro. Splenic arterial dissection and factors leading mutation causing massive spleen infarction. Revista Española De Enfermedades Digestivas 2011;103(6):438–9. doi:10.4321/s1130-01082011000800014.
[14] Nahas Rannie, Pain Thomas, Goulden Peter. A rare cause of epigastric pain in a middle-aged man. Case Rep 2013;2013(1):apr25. doi: 10.1136/bcr-2013-009605.

[15] Neychev Vladimir, Krol Emilia, Dietzek Alan, et al. Unusual presentation and treatment of spontaneous celiac artery dissection. J Vasc Surg 2013;58(2):491–5. doi: 10.1016/j.jvs.2012.10.136.

[16] Nordanstig J, Gerdes H, Kocyz E. Spontaneous isolated dissection of the celiac trunk with rupture of the proximal splenic artery: a case report. Eur J Vasc Endovasc Surg 2009;37(2):194–7. doi: 10.1016/j.ejvs.2008.10.009.

[17] Park Won J, Seo Jung W. Follow-up with computed tomography after spontaneous isolated dissection of the splanchnic artery. Clin Imaging 2018;52:1–7. doi: 10.1016/j.clinimag.2018.01.019.

[18] Sahni Daisy A, Jit Indar B, Gupta CN, Gupta DM, Harjeet E. Branches of the splenic artery and splenic arterial segments. Clin Anat 2003;16(5):371–7. doi: 10.1002/ca.10172.

[19] Shiraki Hiroaki, Kasamoto Manabu, Yasutomi Masamichi, Kaji Shuichiro, Akutsu Koichi, Furukawa Yutaka, et al. Clinical features of spontaneous isolated dissection of abdominal visceral arteries. J Clin Med Res 2020;12(1):13–17. doi: 10.14740/jocmr3916.

[20] Tokue Hiroyuki, Tsushima Yoshito, Endo Keigo. Imaging findings and management of isolated dissection of the visceral arteries. Jpn J Radiol 2009;27(10):430–7. doi: 10.1007/s11604-009-0366-8.

[21] Vancauwenberghe T, Snoeckx A, Vanbeekvoort D, Dymarkowski S, Vanhoenacker FM. Imaging of the spleen: what the clinician needs to know. Singapore Med J 2015;56(3):133–44. doi: 10.11622/smedj.2015040.

[22] Wang Hsiao-Chia, Chen Jiann-Hwa, Hsiao Chieh-Chien, Jeng Chin-Ming, Chen Wei-Lung. Spontaneous dissection of the celiac artery: a case report and literature review. Am J Emerg Med 2013;31(6). doi: 10.1016/j.ajem.2013.02.007.

[23] Woolard Jason D, Ammar Alex D. Spontaneous dissection of the celiac artery: a case report. J Vasc Surg 2007;45(6):1256–8. doi: 10.1016/j.jvs.2007.01.048.

[24] Zafar Yousaif, Meidl Laura, Lonney Maddison, Ullah Waqas, Rashid Mamoon U. Coeliac trunk dissection causing splenic infarction: a comprehensive review. BMJ Case Reports 2019;12(10). doi: 10.1136/bcr-2019-231087.

[25] Zenda Takahiro, Araki Ichiro, Hamano Naomichi, Nishida Hiroto, Ikeda Masatoshi, Bunko Hisashi, et al. Spontaneous celiac and splenic artery dissection. Hiroshima J Med Sci 2017;66(1):21–5.