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Case Report

Spontaneous isolated celiac and splenic artery dissection with splenic infarction ☆

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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.

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.

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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 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.

Case report

A 57-year-old man presented to the emergency department (ED) with 6 hours of acute-onset post-prandial abdominal 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 orchiectomy 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 suspicion 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 period between presentation and follow up imaging. Follow up CT angiogram at 6 months demonstrated marked interval improvement of the dissection with only a very small focal dissection 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

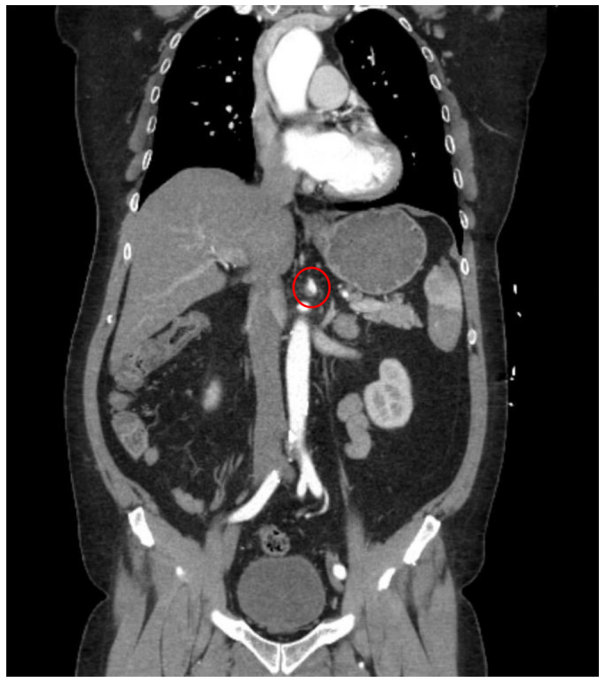

Fig. 1.1 – Coronal CTA demonstrating dissecting celiac artery with thrombosed false lumen.

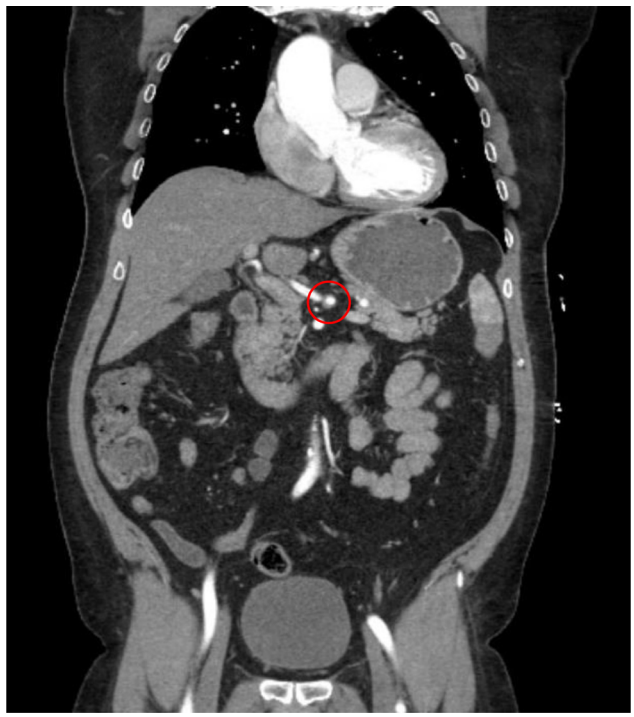

Fig. 1.2 – Coronal CTA demonstrating axial view of dissecting splenic artery with thrombosed false lumen. The intact common hepatic artery is also visualized

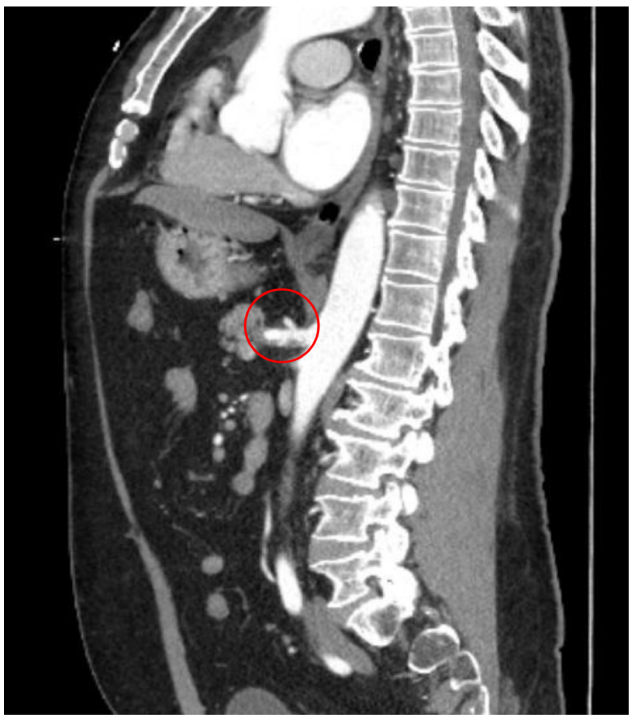

Fig. 1.3 – Sagittal CTA demonstrating dissecting Celiac Artery with contrast flow in false lumen and intact left gastric artery.

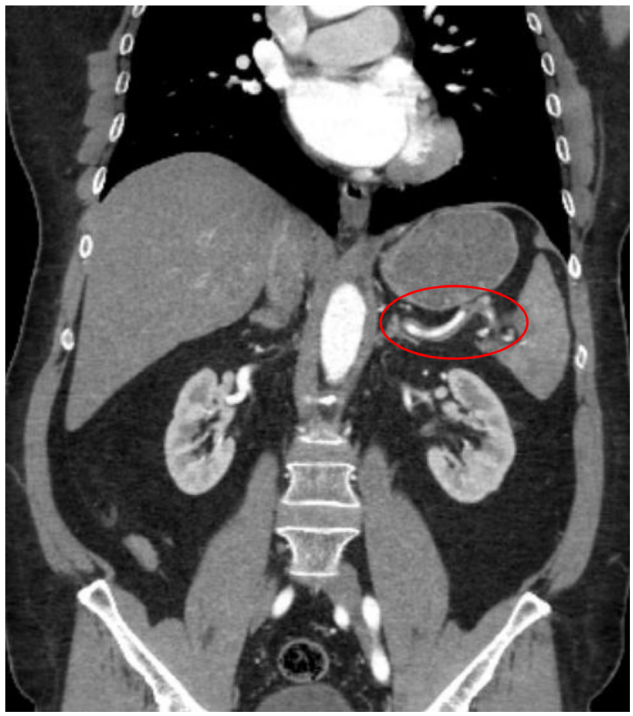

Fig. 1.4 – Coronal CTA demonstrating thrombosed false lumen along length of splenic artery and into the splenic hilum.

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

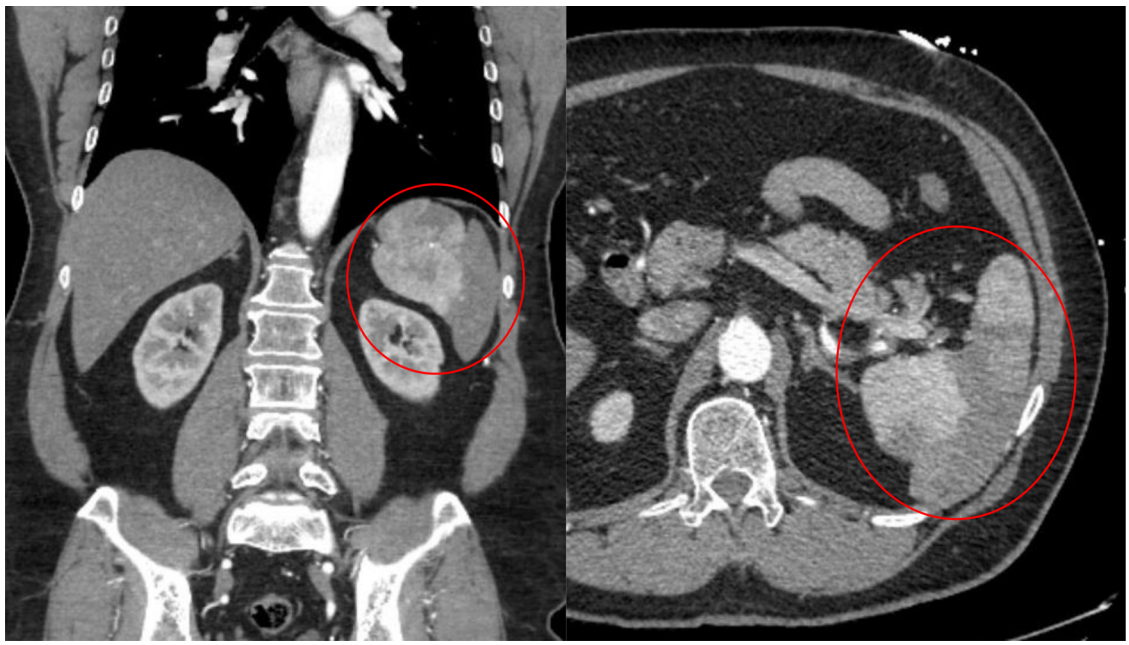

Fig. 1.5 – Coronal and axial CTA demonstrating splenic infarction.

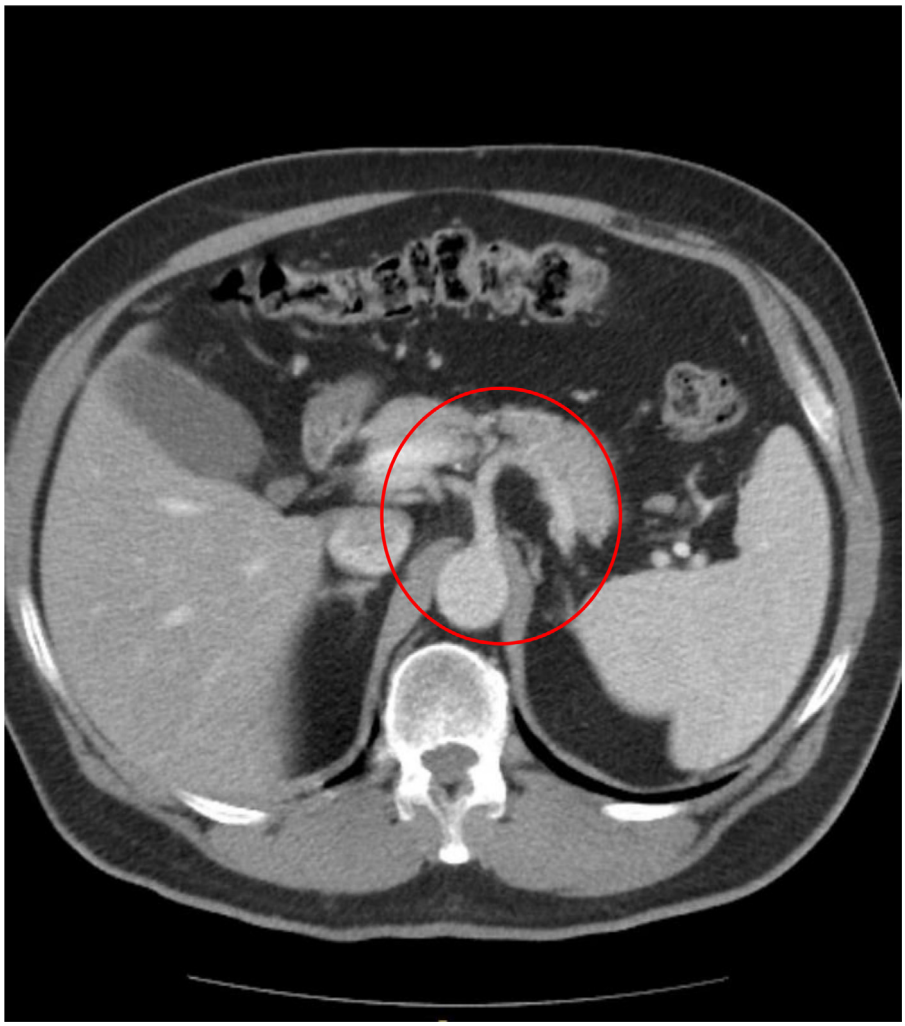

Fig. 2 – CT Chest/Abdomen/Pelvis with contrast performed 3 months prior to arterial dissection demonstrating patent celiac, splenic, and common hepatic arteries with no evidence of dissection.

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-

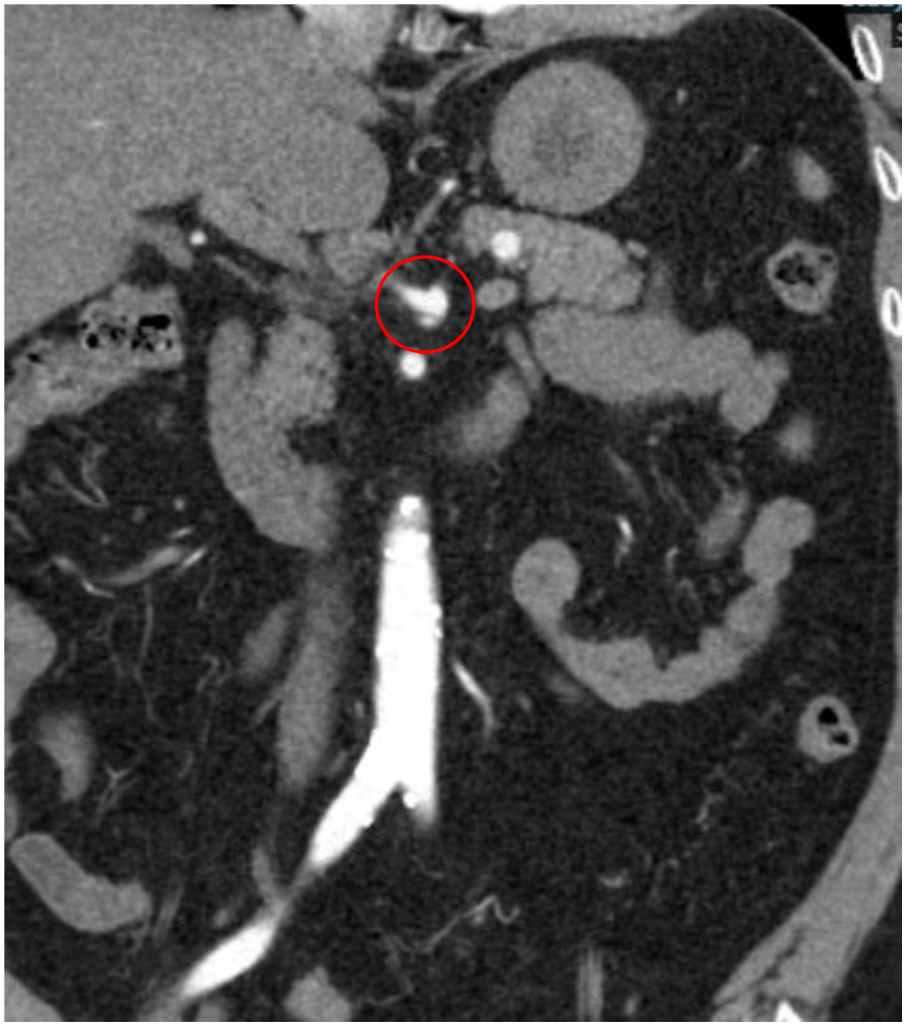

Fig. 3 – CTA abdomen 6 months after dissection demonstrating stable small dissection at distal celiac artery with patent celiac artery branches.

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 regiments 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 patient 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’ digression 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

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 [3,4,4,6,8–10,12,14,15,20,23–25]. Follow up imaging has demonstrated stable dissection and splenic scaring 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 asymptomatically [11,19,20,25]. Remarkably, one patient with spontaneous isolated celiac artery dissection without splenic artery involvement presented asymptomatically 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.

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