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Case Report

Spontaneous Celiac Artery Dissection with Splenic Infarction: A Report of Two Cases

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Spontaneous isolated celiac artery dissection (SICAD) is a rare condition that is characterized by sudden onset abdominal pain, typically occurring in middle-aged men. Although its clinical course is mostly benign, it may progress to true lumen occlusion. No established therapeutic guidelines are available for SICAD associated with splenic infarction. This report describes two patients who presented with sudden onset abdominal pain and were diagnosed with SICAD with splenic infarction based on computed tomography (CT) findings. Patients were treated with bowel rest and anticoagulants. After a week of medical therapy, the abdominal pain resolved. Follow-up CT revealed no progression of the dissection flap. The patients received oral anticoagulants for 3 months and did not experience any symptom recurrence. Medical therapy with anticoagulants may be considered for patients with SICAD and splenic infarction. Associated splenic infarction itself is not an indication for endovascular or surgical intervention for SICAD.

Key Words: Celiac artery, Dissection, Splenic infarction, Anticoagulants

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INTRODUCTION

Spontaneous isolated celiac artery dissection (SICAD) is a rare condition characterized by sudden onset abdominal pain, typically occurring in middle-aged men [1,2]. A recent increase in the use of computed tomography (CT) in patients with abdominal pain has led to an increase in the diagnosis of SICAD [3]. However, the natural history of the condition remains unclear. Although the clinical course is mostly benign, the disease may progress to true lumen occlusion, leading to organ ischemia, aneurysmal dilatation, and rupture [4-6]. Initial medical therapy is the mainstay treatment. Endovascular therapy should be considered for patients with severe persistent symptoms and signs of organ ischemia or rupture [4]. However, no established therapeutic guidelines are available, particularly for celiac artery dissection extending to the branches and associated with splenic infarction or liver dysfunction.

This report presents two cases of SICAD that extended to the splenic and common hepatic arteries and were associated with splenic infarction. Both patients were successfully managed with anticoagulation therapy, without the need for surgical or endovascular interventions. This study was approved by the Institutional Review Board (IRB) of Chung-Ang University Hospital, which waived the informed consent requirement due to minimal patients risks (IRB no. 2301-014-19453).

CASE

1) Case 1

A 56-year-old male presented to the emergency department with epigastric and left upper quadrant pain that had

started the day before his presentation. He experienced sudden onset severe epigastric pain that gradually progressed to the left upper quadrant. He denied having other gastrointestinal symptoms, such as vomiting, diarrhea, melena, or hematochezia. Six years ago, he had undergone subtotal gastrectomy with Billroth II anastomosis for early gastric cancer. He was in excellent general health with no tumor recurrence prior to the onset of his symptoms. The patient denied having any prior episodes of intractable abdominal pain. He had no history of hypertension but had a 25-pack-year smoking history.

The patient was afebrile and hemodynamically stable upon admission, with a blood pressure of 130/70 mmHg and a pulse rate of 76 beats per minute. Physical examination revealed tenderness in the left upper quadrant. Results of the respiratory and cardiovascular examinations were unremarkable. Laboratory tests revealed a mildly elevated white blood cell (WBC) count at 10,890/μL and normal C-reactive protein (CRP) levels at 2.2 mg/L (normal range, 0-5 mg/L). Aspartate transaminase (AST), alanine aminotransferase (ALT), and amylase levels were within normal ranges. Abdominal CT scans revealed celiac artery dissection extending to the common hepatic and splenic arteries, with approximately one-third of the spleen showing signs of infarction. No perfusion defects were observed in the liver (Fig. 1).

The patient was conservatively managed with bowel rest, total parenteral nutrition, and intravenous antibiotics. Anticoagulation therapy was initiated with low molecular weight heparin (LMWH) at a dose of 1 mg per kilogram every 12 hours. On the day following his admission, his body temperature was 38.1°C, returning to normal 3 days thereafter. His WBC count remained elevated for 3 days (13,500 and 15,720/μL) before returning to within the normal range. His CRP levels were elevated up to 14.2 mg/L. His abdominal pain gradually diminished and resolved by the sixth day of admission. Follow-up CT scans performed a week later revealed a stable extent of dissection and decreased intraluminal hematoma. The extent of the splenic infarction slightly increased (Fig. 2). The patient was discharged without fever or abdominal pain 12 days after admission.

After discharge, the patient was prescribed rivaroxaban at a therapeutic dose for 3 months. The elevated CRP levels normalized within 3 weeks of symptom onset. Follow-up CT scans performed 3 months later revealed partial remodeling of the celiac artery dissection, complete remodeling of the common hepatic and splenic arteries, and atrophy of the previously infarcted spleen (Fig. 3). CT scans performed a year later revealed stable focal dissection of the celiac artery. The patient was regularly followed up and experienced no symptom recurrence for 25 months.

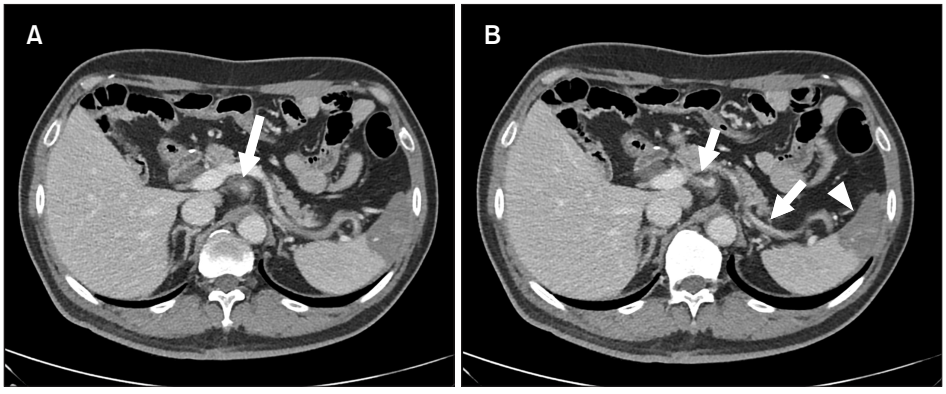

Fig. 1. Computed tomography scan revealed (A) celiac artery dissection with intraluminal hematoma (arrow) and (B) dissection extending to the common hepatic and splenic arteries (arrows) with associated splenic infarction (arrowhead).

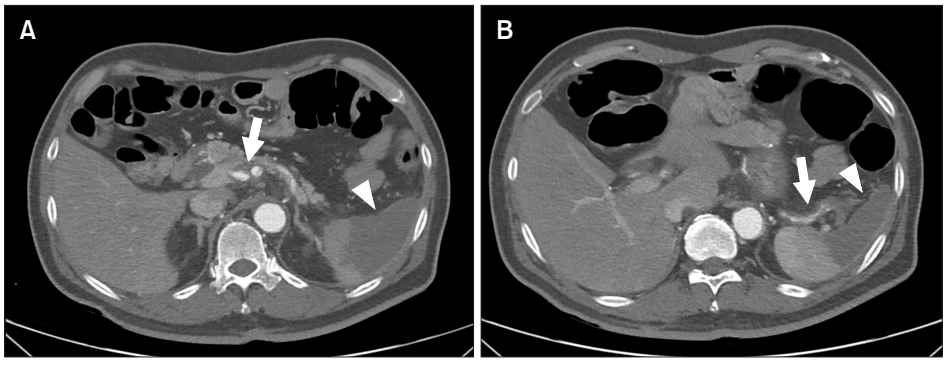

Fig. 2. Computed tomography scan acquired 1 week later revealed (A) no progression of dissection in the celiac, common hepatic, and (B) splenic arteries, along with a decrease in intraluminal hematoma (arrows). The extent of splenic infarction slightly increased (arrowheads).

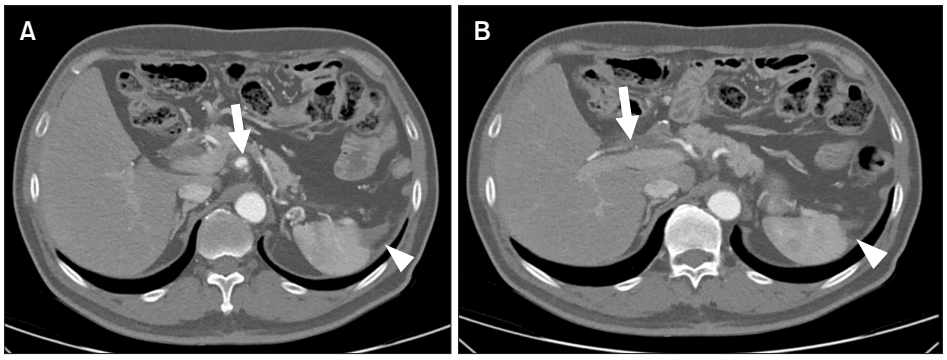

Fig. 3. Computed tomography scan acquired after 3 months revealed (A) partial remodeling of celiac artery dissection (arrow) and (B) complete remodeling in the common hepatic artery (arrow), along with atrophy of the previously infarcted spleen (arrowheads).

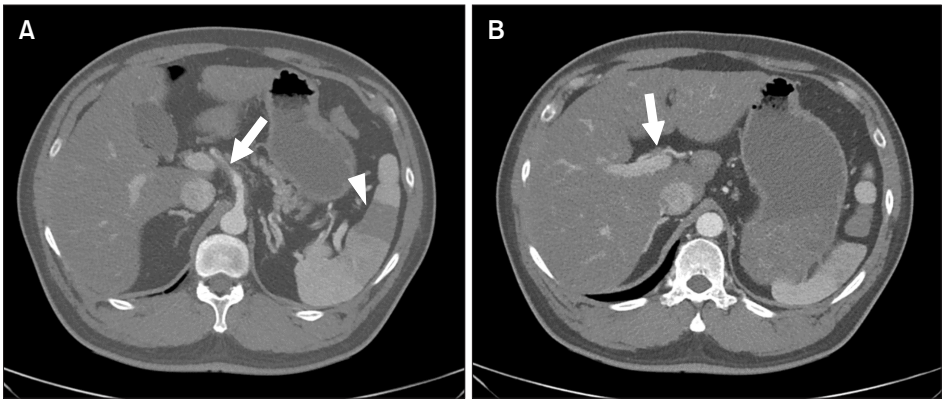

Fig. 4. Computed tomography scan revealed (A) celiac artery dissection with intramural hematoma (arrow) and partial infarction of the spleen (arrowhead) and (B) dissection extending to the right hepatic artery (arrow).

2) Case 2

A 50-year-old male presented to the emergency department with severe intractable epigastric pain that had started suddenly 4 hours beforehand. He did not report any other gastrointestinal symptoms such as vomiting, diarrhea, or melena. He had no history of hypertension and had quit smoking a year prior to his presentation. The patient denied any history of abdominal surgery.

The patient was afebrile and hemodynamically stable upon admission, with a blood pressure of 120/80 mmHg and a pulse rate of 68 beats per minute. Physical examination revealed tenderness in the left upper quadrant. Laboratory tests revealed normal range of WBC (7,340/μL) and CRP level (0.7; normal range, 0-5 mg/L). AST and ALT levels were elevated at 151 IU/L (normal range, 0-34 IU/L) and 171 IU/L (normal range, 0-40 IU/L), respectively; however, these levels were elevated even prior to admission because of the pre-existing fatty liver. Initial abdominal CT revealed celiac artery dissection extending to the common hepatic, proximal right hepatic, and splenic arteries, associated with a partial splenic infarction (Fig. 4).

The patient was conservatively managed with bowel rest, total parenteral nutrition, and intravenous antibiotics. Anticoagulation therapy was initiated with LMWH at a therapeutic dose. On follow-up laboratory tests, the WBC count remained within the normal range; however, the CRP level increased to 16.7 mg/L. After 3 days of bowel rest and parenteral nutrition, abdominal pain resolved. Follow-up CT scans performed 1 week later revealed no progression of the dissection or intraluminal hematoma and a mark-

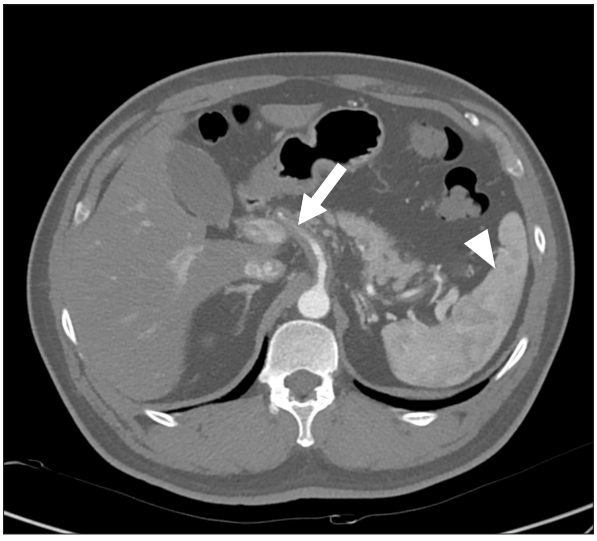

Fig. 5. Computed tomography scan acquired 1 week later revealed the stable extent of celiac artery dissection with an intraluminal hematoma (arrow) and a marked decrease in the extent of splenic infarction (arrowhead).

edly decreased extent of the splenic infarction (Fig. 5). The patient was discharged without fever or abdominal pain 10 days after hospitalization. The patient was prescribed rivaroxaban at a therapeutic dose for 3 months. The patient had no symptom recurrence for 11 months.

DISCUSSION

SICAD is rare and therefore its natural history remains unclear. Superior mesenteric artery (SMA) dissection is the most commonly reported type of mesenteric artery dissection, and conservative managements is widely accepted in cases without bowel ischemia [7]. However, there is no consensus regarding the optimal management of SICAD. Most previous studies have shown that initial conservative management is safe and feasible in stable patients [2,4,8]. However, the prognosis of SICAD is not always benign and can be unpredictable. Cases of proximal splenic artery rupture, aneurysmal changes in the celiac artery, and retroperitoneal hemorrhage have been reported [9,10]. Appropriate patient selection for endovascular or anticoagulation therapy is crucial in the management of SICAD.

Indications for endovascular therapy in SICAD include persistent symptoms despite conservative treatment, severe organ ischemia, increased dissection length and thrombosed lumen, aneurysmal dilation, and rupture [11,12]. In addition, the European Society for Vascular Surgery guidelines recommend considering endovascular revascularization in patients who do not respond to medical management and exhibit suspected bowel ischemia [13]. Several studies have reported the results of endovascular therapy for SICAD. In a study by Kang et al. [11], which included 16 patients with symptomatic SICAD, stent insertion was necessary in 7 patients, and all stents remained patent during a median follow-up of 77 months. Zhou et al. [12] reported that stent insertion was required in 20 of 51 patients, and that the complete remodeling rate of the celiac artery was significantly higher in patients who had undergone endovascular therapy than in those who had undergone conservative management.

However, the use of antiplatelet agents or anticoagulants for the treatment of SICAD remains controversial. Antithrombotics may help decrease the risk of thrombus formation and stenosis in the true lumen. In contrast, they can lead to disease progression by inhibiting the formation of thrombus and increasing the dissection length of the false lumen [14]. A recent case series has recommended conservative management without antithrombotic agents for stable patients [2,4,8]. Regarding the management of SMA dissection, the two largest cohort studies reached a conclusion that antithrombotic therapy did not improve clinical outcomes [15,16]. However, some centers routinely use antiplatelet agents or anticoagulants for all patients. In a systematic review and meta-analysis of 60 studies, 58% of the patients with SICAD did not receive anticoagulants or antiplatelet agents, 21% received anticoagulants, 11% received antiplatelet agents, and 9% received both [1]. Further studies with larger cohorts are needed to define the role of antithrombotic agents, particularly in patients with SICAD.

In the present cases, initial medical therapy including bowel rest, antibiotics, and anticoagulants was administered. Since only several case reports of celiac artery dissection with splenic infarction have been reported, no definite therapeutic guidelines exist for this condition. However, in previous studies, celiac artery dissection and associated splenic infarction were successfully treated with medical therapy, and antiplatelet agents or anticoagulants were used in most cases [17,18]. The spleen is usually supplied by collateral flows along with the splenic artery, which means that surgical or endovascular therapy is not warranted in most cases. In the present cases, although splenic infarction was present, the vital signs were stable, and the abdominal pain resolved; thus, initial conservative management was chosen and continued. Moreover, the first patient had previously undergone subtotal gastrectomy with the remnant stomach supplied by the splenic artery, suggesting that total gastrectomy may be necessary when planning splenectomy. However, both patients recovered well without invasive treatment. Therefore, associated splenic infarction alone does not indicate a need for surgical or endovascular treatment for SICAD.

A long dissection length with branch involvement can be associated with severe true lumen stenosis. Gao et al. [19] demonstrated that the dissection length was an independent risk factor for endovascular treatment of SICAD. In addition, Hau et al. [20] revealed that radiological characteristics, such as branch extension are associated with symptoms. Thus, in-hospital monitoring and follow-up with serial imaging studies are warranted for SICAD patients with branch involvement and splenic infarction, even if conservative management is selected. In addition, follow-up imaging studies after initial therapy are mandatory because late aneurysmal changes or secondary interventions during follow-ups have been reported [8,12]. Consistent recommendations on the frequency of follow-ups remain lacking. Some authors recommend repeated short-term follow-ups with imaging studies within 3 months of the initial episode [4,8,11]. Although short-term follow-up recommendations vary, most studies recommend CT scans at 6 and 12 months, followed by an annual scan until the dissection stabilizes without aneurysmal change [2,4,8,11].

In conclusion, conservative management with anticoagulants can be considered for managing patients with celiac artery dissection presenting with splenic infarction. Associated splenic infarction is not an indication for surgical or endovascular treatment of SICAD. A long dissection length with branch involvement can be associated with severe true lumen stenosis; therefore, careful observation is warranted. Long-term follow-up of large cohort is required to establish treatment guidelines for SICAD, particularly in patients with splenic infarction.

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CONFLICTS OF INTEREST

Suh Min Kim has been the associate editor of VSI since 2022. She was not involved in the review process. Otherwise, no potential conflict of interest relevant to this article was reported.

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AUTHOR CONTRIBUTIONS

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