Acalculous Ischemic Cholecystitis Caused by Spontaneous Celiac Artery Dissection

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Abstract:
We herein report a case of spontaneous isolated dissection of the celiac artery. A Japanese man in his 50s visited an emergency unit, complaining of sudden epigastralgia. Contrast-enhanced computed tomography indicated dissection of the celiac artery with patent false and true lumina, extending to the splenic and common hepatic arteries. On day 3 of hospitalization, the dissection progressed to the proper and right hepatic arteries. Progression of the dissection to the right hepatic artery provoked acalculous ischemic cholecystitis, and cholecystectomy followed. The resected gallbladder revealed extensive aseptic necrosis with little inflammatory reaction, and the gallbladder neck was spared from ischemia.

Key words: acalculous ischemic cholecystitis, spontaneous dissection of celiac artery, contrast-enhanced computed tomography


Introduction
Spontaneous isolated dissection of the celiac artery (SIDCA) was first described by Bauersfeld in 1947 (1). Advances in diagnostic imaging modalities, such as the advent of ultrasound and contrast-enhanced computed tomography (CT), has greatly improved our recognition of SIDCA. However, there are no guidelines concerning the diagnosis or treatment of SIDCA, and no large-scale clinical trials have been performed.

SIDCA often occurs in middle-aged men with a history of smoking, hypertension and hyperlipidemia (2, 3). The major complaint is sudden abdominal pain without guarding or rigidity, but some patients remain asymptomatic. Elevated leukocyte counts and abnormal liver enzyme levels are commonly complicated (3). In a majority (80%) of cases, complete remission is reached with conservative treatment, including anti-hypertensive, anti-platelet and anti-coagulation therapy along with fasting and intravenous infusions (2, 3). Rupture and luminal obstruction of the celiac artery may provoke circulatory insufficiency, thereby necessitating surgical or endovascular treatments (3).

We herein report a middle-aged man with acalculous ischemic cholecystitis caused by SIDCA. The patient had a history of smoking, hypertension and hypercholesterolemia. Dissection of the celiac artery with patent false and true lumina extended to the splenic and common hepatic arteries. At the third day of hospitalization, dissection progressed to the proper and right hepatic arteries, and the occlusion of the right hepatic artery caused extensive ischemic necrosis of the gallbladder.

Case Report
A Japanese man in his 50s visited the emergency unit of Shimada Municipal Hospital, Shimada, Shizuoka, Japan, with a complaint of sudden epigastric pain, radiating to the...
left-sided back and lasting for 30 minutes. The pain happened while extending and stretching his back. He had a history of untreated hypertension and hypercholesterolemia. He had smoked 12 cigarettes/day for 18 years and drunken 1 cup of distilled spirits per day. His family history was unremarkable.

His body temperature was 37.2°C, blood pressure 135/91 mmHg, heart rate 76 beats per minute, respiratory rate 16 per minutes and oxygen saturation 95% while breathing ambient air. His weight was 62.0 kg with a body mass index of 21.2. No heart murmur was auscultated. The abdomen was soft with normal bowel sounds. There was tenderness on palpation on the epigastrium without guarding, rigidity or mass formation. The white blood cell count was elevated at 13,200/μL, and the C-reactive protein (CRP) level in the serum was elevated at 2.0 mg/dL, with D-dimer levels of 0.6 μg/dL. The calculated LDL cholesterol level was 158 mg/dL, HDL cholesterol was 64 mg/dL, and triglyceride level was 92 mg/dL. Other laboratory tests, including the levels of hepatobiliary enzymes, were within reference ranges.

An electrocardiogram showed a normal sinus rhythm with right axis deviation and no evidence of ischemia. The cardiothoracic ratio was 49%, and there was no pulmonary congestion on chest X-ray. Contrast-enhanced computed tomography (CT) in the arterial phase indicated dissection of the celiac artery without abdominal aortic dissection. The dissection extends to the common hepatic artery and splenic artery (A, B, D, E). The false lumen is patent, and the true lumen is not stenotic. Neither aneurysm formation nor rupture is observed. Enhancement effects of RHA are lost on the third day of hospitalization (F, circle). The enhancement effects of RHA are are observed on the first day of hospitalization (C, circle).

Fasting was continued, and oral intake of water, including jelly beverages, was started on the third day of hospitalization without exacerbation of abdominal symptoms. Soon af-
After drinking a jelly beverage, he complained of accelerated abdominal pain. There was tenderness at the epigastric and right subcostal regions with guarding and rigidity. The white blood cell count was elevated to 14,300/μL, and the serum CRP level had increased to 16.3 mg/dL, but the serum levels of hepatobiliary enzymes were not elevated. Contrast-enhanced CT showed that the dissection had progressed to the proper and right hepatic arteries in association with obstruction of the right hepatic artery, but neither aneurysm formation, rupture nor obstruction of the true lumen of the celiac, splenic and common hepatic arteries was seen (Fig. 1D-F, 2C). Dilatation of the gallbladder lumen was evident, and loss of enhancement effects of the gallbladder wall was noted (Fig. 2D). No gallstones were recognized.

Emergency laparoscopic cholecystectomy was performed under the diagnosis of acalculous ischemic cholecystitis. Grossly, the surgical material displayed extensive necrosis throughout the gallbladder wall (Fig. 3A). The neck region of the gallbladder remained intact without ischemic changes. Microscopically, the body of the gallbladder showed transmural ischemic necrosis without infiltration of inflammatory cells (Fig. 3B, C). The intact gallbladder neck region was clearly demarcated from the ischemic gallbladder body. No abnormality was noted in the mural arterial branches.

Abdominal pain disappeared five days after cholecystectomy. At the postoperative day 6, contrast-enhanced CT revealed that the dissection of the celiac, splenic and common hepatic artery remained stable, and the right hepatic artery was reopened (Fig. 4). The patient was discharged 16 days after cholecystectomy without complications. The patient has been followed up with anti-hypertensive medication. The celiac arterial dissection has not recurred for more than six years.

**Discussion**

We herein report a case of acalculous ischemic cholecystitis caused by SIDCA. Stimulated gastrointestinal motility...
and accelerated bile excretion facilitated by drinking jelly beverage on the third day of hospitalization might have induced increased oxygen consumption and intraluminal pressure of the gallbladder, provoking acalculous ischemic necrosis of the gallbladder. No gallstones were detected by contrast-enhanced CT or in the surgical specimen. Acalculous ischemic cholecystitis caused by SIDCA has been described in previous case reports (4). Acalculous ischemic cholecystitis has also been recorded in cases of acute aortic dissection (5-8). Acalculous cholecystitis is seen in 5%-14% of acute cholecystitis cases, while acute cholecystitis accompanied gallstones in around 90% of cases (9).

Acalculous cholecystitis may be caused by dysmotility of the gallbladder that increases the intracholecystic pressure to induce ischemia, inflammation and overgrowth of bacteria (10). Dysfunction of gallbladder contraction may result from diabetes mellitus, hypertension, heart failure, renal failure, vasculitis, embolism, acquired immunodeficiency syndrome, infection, surgery, trauma, burn and parental nutrition (10). The reduction in the blood supply from the celiac artery caused by median arcuate ligament syndrome and atherosclerosis is insufficient for inducing ischemic changes in the abdominal organs, such as the stomach, pancreas, spleen, liver and gallbladder, due to the abundance of collateral blood flow (11-14). The pancreatic arcade anastomosing the gastroduodenal artery with the first jejunal artery is the most important blood supply for abdominal organs in cases of celiac arterial obstruction (12-14). In localized celiac artery dissection, the blood flow of the proper hepatic artery is maintained through the pancreatic arcade. The cholecystic artery, typically branching from the anterior right hepatic artery, comprises the main vessel feeding the gallbladder, which also receives numerous arterial twigs from the right hepatic artery via the gallbladder bed on the liver side (15). In the present case, the dissection of the celiac artery progressed to the right hepatic artery, hampering the blood supply to the cholecystic artery, which was not recovered through the pancreatic arcade. Of note, the transmural ischemic changes in the body of the gallbladder showed a clear border with the normal-looking (non-ischemic) mucosa located at the neck of the gallbladder. The present findings indicate that the gallbladder neck had a collateral blood supply in the present case. The pattern of vascular supply to the biliary tract and neck region of the gallbladder reportedly show some degree of variation (16).

It has been suggested that a rapid increase in the arterial pressure after intense exercise may be a causative factor for SICDA, since SICDA occurred during weight-lifting or...
weight training in three cases (17-19). Mechanical stress may cause dissection of the vertebral artery (20). In the present case, epigastric pain occurred during extending and stretching movements of the patient’s back, which might have induced mechanical stress on the celiac artery, thereby provoking SICDA.

Causatively, SIDCA may be correlated with fibromuscular dysplasia, Marfan syndrome, Ehlers-Danlos syndrome, vasculitis syndrome, arteriosclerosis and pregnancy (21-23). Segmental arterial mediolysis (SAM) should also be considered as a potential cause of SICDA (22, 23). In 1976, Slavin and Gonzalez-Vitale (24) first reported three autopsy cases of SAM that had formed in the abdominal muscular arteries. The medial smooth muscle layer revealed segmental necrosis, typically causing intraabdominal hemorrhaging. In an animal model, α1 or β2 adrenergic agonists accelerated the release of noradrenaline from the sympathetic nerve, which induced the apoptosis of smooth muscle cells of the tunica media (25, 26). The outer arterial medial layer resulted in vacuolization and lysis, representative microscopic features of SAM. The most commonly affected vessels are the superior mesenteric (53%), hepatic (45%), celiac (36%), renal (26%) and splenic (25%) arteries, and SAM results in aneurysm formation (76%), dissection (61%) and arterial rupture (46%) (27). Risk factors for SAM include an older age, hypertension and smoking, all of which are common with SIDCA (3, 27). Uchiyama et al. (28) proposed clinical criteria of SMA, including a sudden onset of intraabdominal bleeding, middle to elderly patient age, a lack of vasculitis or arteriosclerosis and beading and narrowing signs on angiography. In the present case, a histopathological evaluation of the involved artery was not performed, so the possibility of celiac arterial dissection secondary to SAM cannot be excluded.

Abnormal serum levels of liver enzymes are reportedly seen in 25% (5/20) of patients with SICDA (3). In the present case, the hepatobiliary enzyme levels were within the reference ranges before and after acalculous ischemic cholecystitis. In the liver, 70% of the blood supply is maintained by the portal vein and 30% by the hepatic artery. The left and right hepatic arteries communicate with each other in the hilar plate (29). Occlusion of the right hepatic artery might have been rescued by the left hepatic artery in the present case.

Arterial dissection can be restricted to the celiac artery, or it may extend from the celiac artery to the splenic and/or
proper hepatic arteries. Kim et al. (2) reviewed a total of 164 SIDCA patients and evaluated the risk of surgical treatments. Aneurysm formation and extended dissection to the arterial branches were associated with an increased risk of invasive treatment. In the present case, the dissection extended to the splenic and common hepatic arteries, thereby assigning the patient a greater risk of invasive surgery.

Conservative treatments are sufficient to manage SICDA in most cases (2, 3), but careful monitoring of the dissection of the celiac artery is necessary, as gastrointestinal motility induced by oral intake might accelerate the extended dissection to the right hepatic artery, thus causing acalculous ischemic cholecystitis. Further studies are needed to clarify the pathological mechanisms and therapeutic strategies of SIDCA.

The authors state that they have no Conflict of Interest (COI).

References


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