CASE REPORT
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A Ruptured Cystic Artery Pseudoaneurysm with Concurrent Cholecystoduodenal Fistula: A Case Report and Literature Review

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Pseudoaneurysms of the cystic artery and cholecystoduodenal fistula formation are rare complications of cholecystitis and either may result from an inflammatory process in the abdomen. A 68-year-old man admitted with acute cholecystitis subsequently developed massive upper gastrointestinal (GI) bleeding. Abdominal computed tomography showed acute calculous cholecystitis and hemobilia secondary to bleeding from the cystic artery. Angiography suggested a ruptured pseudoaneurysm of the cystic artery. Upper GI endoscopy showed a deep active ulcer with an opening that was suspected to be that of a fistula at the duodenal bulb. The patient was managed successfully with multimodality treatment that included embolization followed by elective laparoscopic cholecystectomy. Presently, there is no clear consensus regarding the clinical management of this disease. We have been able to confirm various clinical features, diagnoses, and treatments of this disease through a literature review. A multidisciplinary approach through interagency/interdepartmental collaboration is necessary for better management of this disease. **(Korean J *Helicobacter* Up Gastrointest Res 2018;18:135-141)**

Key Words: Angiography; Cholecystoduodenal fistula; Pseudoaneurysm

INTRODUCTION

Gastrointestinal bleeding from the biliary tree, termed "hemobilia," is an uncommon event. A cystic artery pseudoaneurysm is a very rare complication of cholecystitis and is known to cause hemobilia.[1] Cholecystoenteric fistula is also a rare complication of gallstone disease.[1] We report here a very rare case of upper gastrointestinal hemorrhage due to rupture of pseudoaneurysm of the cystic artery with concurrent cholecystoduodenal fistula.

CASE REPORT

A 68-year-old man presented to the emergency room complaining of epigastric pain. He had a history of diabetes, hypertension and GB stones. Until shortly before his presentation, the patient was taking aspirin and clopidogrel and denied any history of trauma or recent surgery. On clinical examination, he was afebrile and hemodynamically stable. The general physical examination was normal, but there was tenderness and rebound tenderness in the right hypochondrium and epigastrium. The laboratory test results included a white blood cell count of 5,760/mm^3 (normal range, 4,000～8,000/mm^3), a hemoglobin level of 10.9 g/dL (normal range, 13.0～18.0 g/dL), and platelet count of 239,000/mm^3 (normal range, 140,000～450,000/mm^3). Blood chemistry showed the following abnormal values: total bilirubin 3.32 mg/dL (normal range, 0.2～1 mg/dL), aspartate aminotransferase 298 U/L (normal range, 10～40 U/L), alanine aminotransferase 184 U/L (normal range, 10～40 U/L), gamma-glutamyltransferase 714 U/L (normal range, 5～40 U/L), and C-reactive protein 20.8 mg/L (normal range, 0～5 mg/L). The patient was suspected to have an acute cholecystitis or cholangitis because of the history of GB stones and right upper quadrant abdominal pain. Contrast enhanced computed tomography (CT) was planned for confirmation. After CT imaging, the patient suddenly developed massive hematemesis. His hemoglobin dropped to 6.0 g/dL, blood pressure decreased to 50/30 mmHg and his heart rate was 120 bpm at that time. The CT showed a severely

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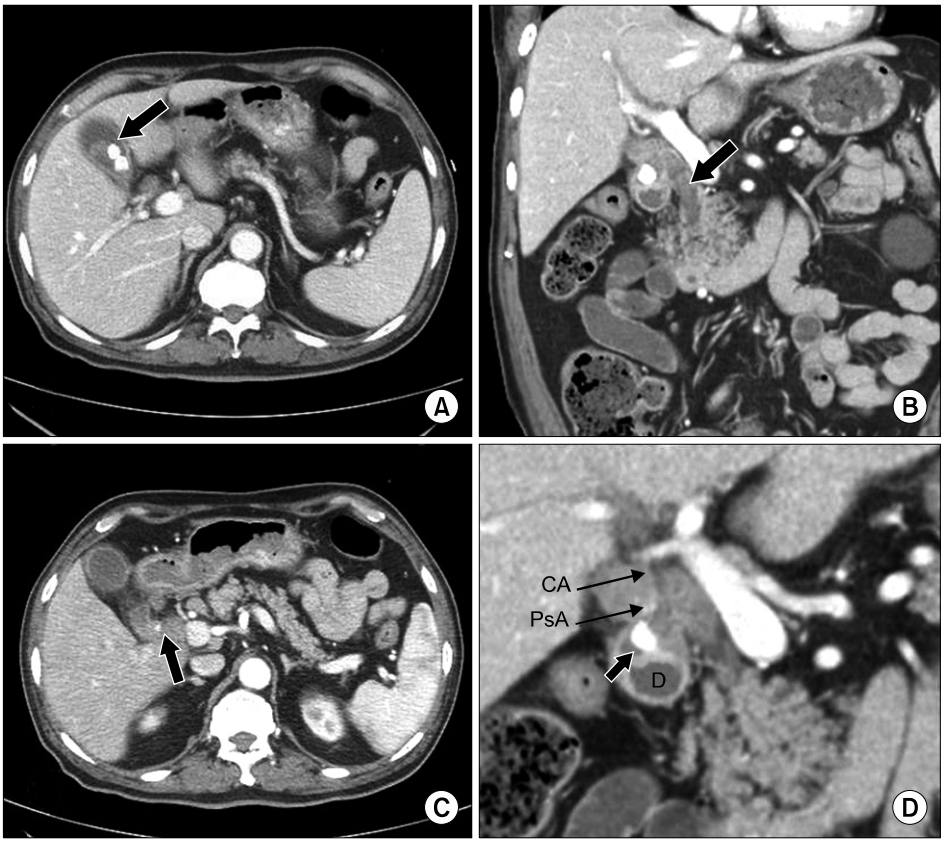

Fig. 1. Abdominal computed tomography (CT) findings. Contrast-enhanced CT images show acute calculous cholecystitis with intragallbladder (GB) hemorrhage (arrow) (A), high density material (arrow) filling the common bile duct (hemobilia) (B), a small-sized pseudoaneurysm of the cystic artery (arrow) (C), and a fistulous tract between the GB and the duodenum (cholecystoduodenal fistula) with a stone (arrow) adjacent to the second portion of the duodenum (D). CA, cystic artery; PsA, pseudoaneurysm; D, duodenum.

distended GB with two 10 mm sized-stones and hemobilia due to cystic artery bleeding. In addition, a fistula tract caused by the pressure of a stone adjacent to second portion of duodenum was suspected (Fig. 1).

We assessed the patient's status as hypovolemic shock due to upper GI bleeding. He underwent an emergency angiography to find the bleeding focus after resuscitation with a massive transfusion (14 units of packed red blood cells). Angiography revealed rupture of the small cystic artery PsA originating from the hepatic artery and a suspicion of extravasation of contrast into duodenum (Fig. 2). A Coil embolization was performed to control the bleeding. He then underwent an upper GI endoscopy which showed that the esophagus and stomach were normal and revealed a deep active ulcer (15 mm) with an opening that was suspected of a fistula formation seen on the anterior inferior surface of the first part of the duodenum (Fig. 3). The patient underwent a follow-up CT scan before surgery. Stones within the GB were found, and the impacted stone in the fistula tract, which was found at

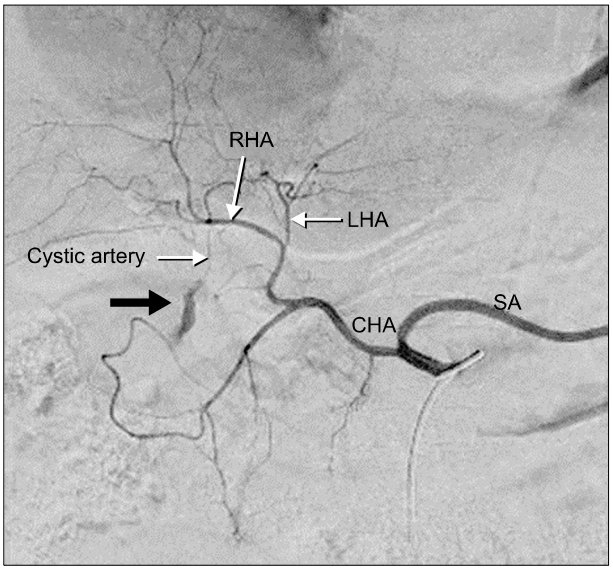

Fig. 2. Angiographic findings. Angiographic images show extravasation from pseudoaneurysm originating from the cystic artery (black arrow). RHA, right hepatic artery; LHA, left hepatic artery; CHA, common hepatic artery; SA, splenic artery.

the time of diagnosis, was found to be released (Fig. 4). After a few days, the patient underwent an elective laparoscopic cholecystectomy. During the operation, there was severe inflammation and fibrosis within the cystic duct and GB infundibulum, and a severe adhesion was observed between the GB and duodenum. After ligating the cystic artery and the cystic duct with a clip, the GB was separated from the liver bed, and then the primary closure of the cystic duct stump and duodenum was performed. Histologically, the GB showed focal wall thinning with an erosive mucosal surface, suspicious for the clinical diagnosis of a cholecystoduodenal fistula (Fig. 5). The patient was discharged without complication on the seventh postoperative day.

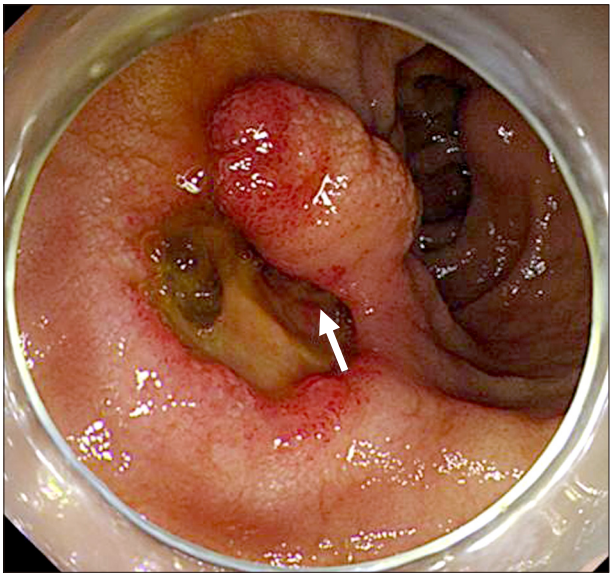

Fig. 3. Endoscopic findings. Endoscopy shows an active duodenal ulcer with an opening (white arrow) that is suspected to be the opening of a fistula located at the duodenal bulb.

DISCUSSION

A PsA of the cystic artery is a very rare cause of hemobilia, and its pathogenesis is unclear. Hemobilia generally presents as upper quadrant pain (biliary colic), obstructive jaundice, and GI hemorrhage (Quincke's triad). Such aneurysms have been thought to occur after inflammatory processes near the vessel and can rupture into the GB, cystic duct or bile duct with resultant hemobilia but rarely rupture into the peritoneal cavity. Most cases reported have been caused by acute calculous cholecystitis except for postoperative complications. Aneurysmal formation can further progress from patient factors such as atherosclerosis, hypertension, bleeding disorder, and vasculitis.[1] PsA of the cystic artery due to acute cholecystitis is a very rare cause of upper GI bleeding. In a PubMed search of the literature (using key words: pseudoaneurysm, cystic artery, and cholecystitis), only 36 cases have been identified in the English literature. Among these cases, only 27 cases of a cystic artery aneurysm were expressed as a clinical manifestation of a rupture such as GI bleeding and internal hemorrhage.

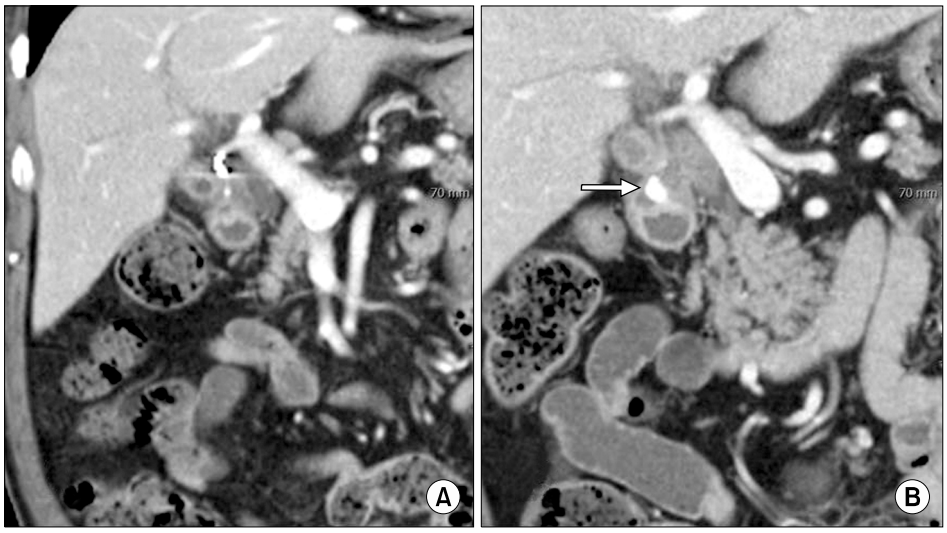

Fig. 4. Follow-up computed tomography (CT) findings. Follow-up CT images show that the impacted stone (arrow) in the fistula tract has been disimpacted (A) compared with an initial CT scan (B).

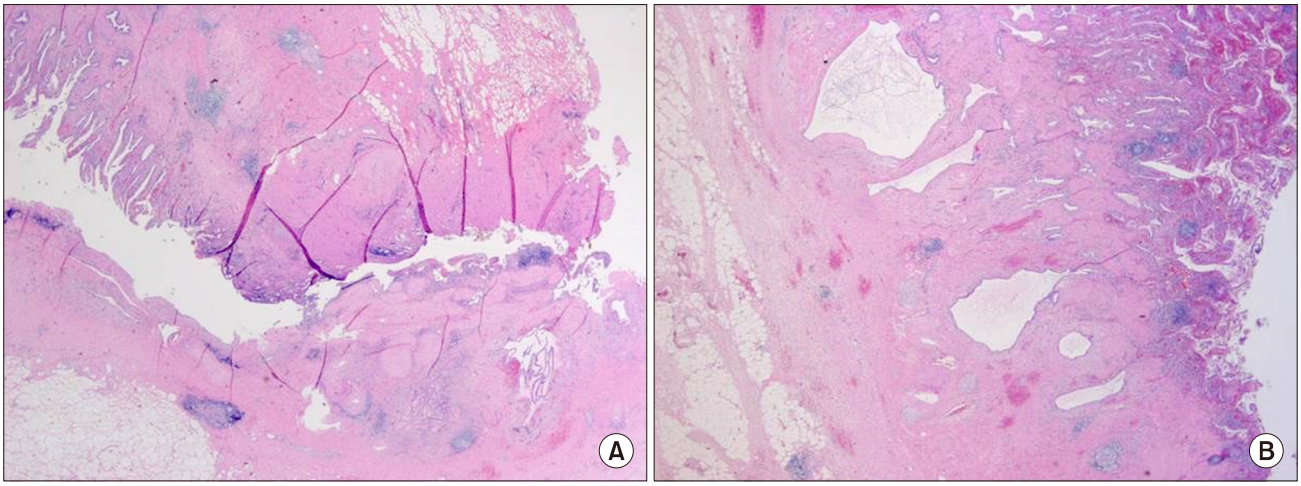

Fig. 5. Histopathological findings (H&E, ×40). The gallbladder shows an erosive mucosal surface with chronic inflammation and wall thinning (A) and epithelial proliferation, muscular hypertrophy and intramural diverticula (Rokitansky-Aschoff sinuses) (B).

We reviewed 36 reports on PsA of the cystic artery related to cholecystitis and the results are as follows (Table 1).[1-30] The mean age of the patients was 67 years, and the sex distribution was 10 males and 26 females. PsA of the cystic artery was found to be 25.0% (9/36) without rupture, and 75.0% (27/36) with rupture. In another report,[2] hemobilia was reported in 45% of cases in the PsA of the cystic artery, but in 75.0% (27/36) of cases in this review. GI manifestations of these hemobilia patients were 46.2% (12/26) for melena, 23.1% (6/26) for hematemesis and 7.7% (2/26) for hematochezia. Nine of the patients showed no symptoms of GI bleeding. In one case,[3] rupture of the PsA of the cystic artery resulted in hemoperitoneum with GB perforation, so there were no hemobilia. The incidence of jaundice was 27.8% (10/36) and the typical three symptoms of hemobilia (Quincke's triad) were reported in 16.7% (6/36) of the cases reported in this review, compared with 32% to 40% reported in other literature.[31] CT or CT angiography was most frequently used for diagnostic purposes, followed by angiography (11.1%, 4/36), Ultrasonography (US) (8.3%, 3/36) and magnetic resonance imaging/magnetic resonance cholangiopancreatography (8.3%, 3/36), and there was no case in which the PsA of the cystic artery was diagnosed by ultrasound (US) alone. The US value can identify potential aneurysms, but it may not detect small aneurysm. Although celiac or selective hepatic arteriography is the gold standard method for diagnosis, a contrast enhanced CT is the best noninvasive method especially in emergency situations such as acute abdominal bleeding.[2,4] In this case, rupture of cystic artery PsA was suspected on CT, and subsequent angiography was able to confirm and treat, simultaneously.

Transcatheter arterial embolization (TAE) followed by cholecystectomy was performed in most cases (41.7%, 15/36). Patients were treated with TAE alone if they were not suitable for surgical candidates due to age or underlying disease, and 25.0% (9/36) cases were treated with surgery alone. There were two cases of an emergency operation due to failure of TAE.[5,6] Traditionally, surgical ligation has been known to treat this disease. Based on the case reviews, the combination of TAE followed by surgical treatment seems to be the most effective. Recently, embolization of the cystic artery has been reported as effected, with a lower risk of visceral trauma and without a need for general anesthesia.[7] Like most other cases, we preformed embolization first, followed by an elective laparoscopic cholecystectomy.

In addition, it is a peculiar point that the cystic artery PsA rupture was presented with a cholecystoduodenal fistula in this patient. The creation of a bilioenteric fistula is a very rare complication of GB stones which affects less than 1% of patients.[32] The fistula can occur anywhere in the GI tract with the most common location being chol-

Table 1. Summary of Cases of Cystic Artery Aneurysm Secondary to Acute Cholecystitis

No.	Year	1st author	Age (yr)	Sex	Presentation	HB	Rup	Diagnosis	Treatment
1	2017	Tapnio RH[7]	91	F	Pain, fever	Y	Y	CT, US	TAE+cholecystectomy
2	2017	Tapnio RH[7]	61	M	Pain, fever	Y	Y	CTA	TAE+cholecystectomy
3	2017	Tapnio RH[7]	91	M	None	N	N	CT	TAE
4	2017	Zucker B[9]	56	M	None	N	N		Cholecystectomy
5	2017	Trombatore C[10]	64	M	Pain, vomiting, hematemesis, melena	Y	Y	CT	TAE+cholecystectomy
6	2016	Alis D[11]	36	M	Pain	N	N	CT	TAE
7	2015	Loizides S[12]	61	F	Pain, vomiting	N	N	CT	Cholecystectomy
8	2015	Muñoz-Villafranca C[13]	74	M	Pain, melena	Y	Y	CT, MRI	TAE
9	2014	Glaysher MA[1]	86	M	Pain, vomiting, melena	Y	Y	CT	Cholecystectomy
10	2014	Kulkarni V[14]	55	M	Pain, jaundice, melena	Y	Y	CT, MRCP	TAE+cholecystectomy
11	2013	Nana GR[15]	74	M	Pain, jaundice, hematemesis	Y	Y	CTA	TAE+cholecystectomy
12	2013	Nana GR[15]	79	F	Weight loss, melena, jaundice	Y	Y	CT	TAE
13	2013	Suzuki S[16]	85	F	Pain, jaundice, Mirizzi syndrome	N	N	CTA, US	TAE+cholecystectomy
14	2013	Fung AK[3]	64	M	Pain	N	Y	CTA	Cholecystectomy
15	2013	Priya H[5]	22	M	Pain, jaundice, melena, hematemesis	Y	Y	Angiography	Cholecystectomy
16	2012	Chong JJ[17]	56	M	Pain, fever, hematemesis	Y	Y	CT	TAE+cholecystectomy
17	2011	Siddiqui NA[18]	58	M	Pain, jaundice	Y	Y	CT	TAE+cholecystectomy
18	2010	Ahmed I[19]	54	M	Pain	Y	Y	CT	TAE+cholecystectomy
19	2010	Hague J[20]	83	M	Pain	Y	Y	CT	TAE
20	2010	Hague J[20]	79	M	Pain	Y	Y	CTA	TAE+cholecystectomy
21	2010	Hague J[20]	83	M	Pain, melena	Y	Y	CTA	TAE
22	2010	Nkwam N[21]	71	M	Pain, vomiting	N	N	CTA	Cholecystectomy
23	2010	Desai AU[22]	78	F	Pain, vomiting, melena	Y	Y	CT	TAE
24	2009	Mullen R[23]	75	F	Pain, melena	Y	Y	CT	TAE
25	2009	Mullen R[23]	82	M	Pain	N	N	CT	TAE
26	2008	Machida H[4]	71	M	Pain	N	N	CT	Cholecystectomy
27	2008	Shimada K[24]	68	M	Jaundice	Y	Y	CT	Hepatectomy
28	2007	Akatsu T[25]	58	M	Pain, jaundice	Y	Y	MRI	Cholecystectomy
29	2007	Saluja SS[26]	43	F	Melena, hematemesis	Y	Y	CT, US	TAE+cholecystectomy
30	2006	Pérez JL[27]	77	F	Hypotension, anemia	Y	Y	Angiography	TAE
31	2006	Lee JW[8]	72	F	Pain, vomiting, hematochezia	Y	Y	CT	TAE+cholecystectomy
32	2006	Joyce MR[28]	58	M	Pyrexia, melena	Y	Y	Explore laparotomy	Cholecystectomy
33	2004	Gutiérrez G[6]	66	F	Pain, anemia, hematochezia	Y	Y	CT	Cholecystectomy
34	2002	Maeda A[29]	62	M	Pain	N	N	Angiography	TAE+cholecystectomy
35	1999	Delgadillo X[2]	28	M	Pain, jaundice, hematemesis	Y	Y	CT	TAE
36	1996	Nakajima M[30]	72	M	Pain, jaundice, melena	Y	Y	CT, angiography	Cholecystectomy

HB, hemobilia; Rup, rupture; F, female; M, male; Y, yes; N, no; CT, computed tomography; US, ultrasonography; CTA, CT angiography; MRI, magnetic resonance imaging; MRCP, magnetic resonance cholangiopancreatography; TAE, transcatheter arterial embolization.

ecystoduodenal (~60%) and cholecystocolic (17%) and cholecystogastric and choledochoduodenal areas (5%).[33] The incidence of a cholecystoduodenal fistula is reported to occur during 0.29% to 0.42% of cholecystectomy procedures.[34,35] The most common cause of cholecystoduodenal fistula is a gallstone.[36] A fistula is often diagnosed during surgery. However, there are several cases[34] that were diagnosed before surgery due to symptoms of gallstone ileus and cholangitis. The classic treatment of a cholecystoenteric fistula is cholecystectomy with a primary closure of the fistula.[34] Except our case, there was only one case report[5] with two rare complications of cholecystitis that occurred simultaneously worldwide. In 36 case reviews, fistulas were found only in 3 cases, and the types of fistulas included cholecystoduodenal fistula,[5] cholecystojejunal fistula[1] and cholecystocolonic fistula.[8]

Even though a PsA of the cystic artery and cholecystoenteric fistula are very rare, they should be considered as a complication of calculous cholecystitis, and a PsA of the cystic artery should be included in the differential diagnosis of hemobilia. There is no clear consensus yet on the clinical management of this disease. We have been able to confirm various clinical features, diagnoses, and treatments of this disease through a literature review. A CT scan and angiography are useful for diagnosis, and TAE followed by cholecystectomy seems to be best for treatment of this disease. Additionally, TAE alone therapy is also considered an effective temporary option in the subset of these patients who are unable to undergo immediate surgical management. A multidisciplinary collaboration between radiologists and surgeons is the key-point to improve management of these patients.

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