



Case Report

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화농성 척추추간판염 환자에서 발생한 비결석성 출혈성 담낭염 1예

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Hemorrhagic Acalculous Cholecystitis in Pyogenic Spondylodiscitis Patient

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Hemorrhagic cholecystitis (HC) is a rare form of cholecystitis that may be caused by the progression of either calculous or acalculous cholecystitis, which has been reported sporadically as case reports. Acalculous cholecystitis is often diagnosed late and can be fulminant. HC without gallstones also can have a poor prognosis and be linked to a high mortality rate. Therefore, early detection, hemodynamic monitoring, and prompt treatment are essential to treat bleeding in cholecystitis. We report a case of hemorrhagic acalculous cholecystitis discovered during the examination and point-of-care ultrasound in a patient with pyogenic spondylodiscitis. The patient was referred with a chief complaint of liver dysfunction without bleeding tendencies or risk factors. Ultrasound revealed focal gallbladder wall irregularity, intraluminal membranes, and non-shadowing non-layering intraluminal echoes. Extravascular contrast leakage was exhibited on the computed tomography scan. The patient underwent emergency percutaneous drainage and subsequent cholecystectomy before developing major complications.

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INTRODUCTION

Hemorrhagic cholecystitis (HC) is a relatively uncommon type of cholecystitis that can occur as a result of the advancement of either calculous or acalculous cholecystitis. This condition has been infrequently reported in the medical literature and is typically described through sporadic case reports. Conventionally, most

cases are caused by gallstones and have been associated with factors such as trauma, vascular anomaly, neoplasms of the biliary system, and parasite infection.¹ Spontaneous HC has also been reported in patients with a higher tendency for bleeding, such as those with uremia-induced bleeding in dialysis patients² or in patients using antiplatelet or anticoagulant drugs.³⁻⁵ Early detection, hemodynamic monitoring, and timely treatment are essential to

treat bleeding in cholecystitis.^{3,6} We report a case of hemorrhagic acalculous cholecystitis discovered during the examination and point-of-care ultrasound in a patient who presented with a chief complaint of elevated liver enzyme without the previously mentioned bleeding tendencies or risk factors. The Institutional Review Board of Keimyung University Dongsan Hospital approved this study (IRB No. 2022-12-074).

CASE

A 66-year-old woman presented to the tertiary hospital with chief complaints of low back pain that had persisted for 2 weeks, chills, general weakness, vomiting, and an altered level of consciousness. The patient had undergone a hysterectomy and bilateral salpingo-oophorectomy for endometrial cancer several decades ago, was a carrier of immune-inactive phase chronic hepatitis B and had undergone spinal fusion for L1–2 lumbar disc herniation and L1–5 lumbar spinal stenosis 7 years previously. The patient had denied history of antiplatelet or anticoagulant use. Chest computed tomography (CT) at the emergency department showed aspiration pneumonia, while lumbar magnetic resonance imaging (MRI) showed findings suggesting L2–3 pyogenic discitis. Gram-positive cocci and *Streptococcus agalactiae*, respectively, were identified in the peripheral blood and dorsal soft tissue abscess cultures performed at a previous hospital at the time of transfer. Upon the diagnosis of pyogenic spondylodiscitis with

psosas infiltration and aspiration pneumonia, endotracheal intubation and ceftriaxone and clindamycin therapies were initiated. After 10 days of mechanical ventilation in the intensive care unit, the patient was transferred to the general ward unit and showed stable vital signs without fever while placed on tube feeding. CT performed on hospital day (HD) 21 showed improvements in the pneumonia lesions, so the patient was transitioned to a soft diet. On HD 28, a gastroenterology consult was requested due to abnormal liver function test (LFT) findings. At the time of consult, her vital signs included a blood pressure of 143/85 mmHg, a pulse of 99 beats/min, a body temperature of 37.2 °C, an oxygen saturation of 98%, and the following blood test parameters: white blood cell count 9,020/ μ L, hemoglobin 9.2 g/dL, platelet count 264,000/ μ L, Na-K-Cl 136-3.8-102 mmol/L, blood urea nitrogen/creatinine 6/0.28 mg/dL, total protein and albumin 6.0/2.8 g/dL, alkaline phosphatase/aspartate transaminase/alanine transaminase 962/278/340 U/L, total/direct bilirubin 4.25/3.86 mg/dL, gamma-glutamyl transferase 309 IU/L, prothrombin time 13.5 seconds (international normalized ratio 1.18), and C-reactive protein 6.2 mg/dL (reference 0.0–0.5). The patient was on ceftriaxone monotherapy then, and no non-steroidal anti-inflammatory drugs had been prescribed since the time of referral. The latest LFT performed 2 days before the referral did not show any abnormalities. At the time of examination, the patient was bedridden with severe general weakness, did not express any symptoms, including abdominal pain, and did not show any

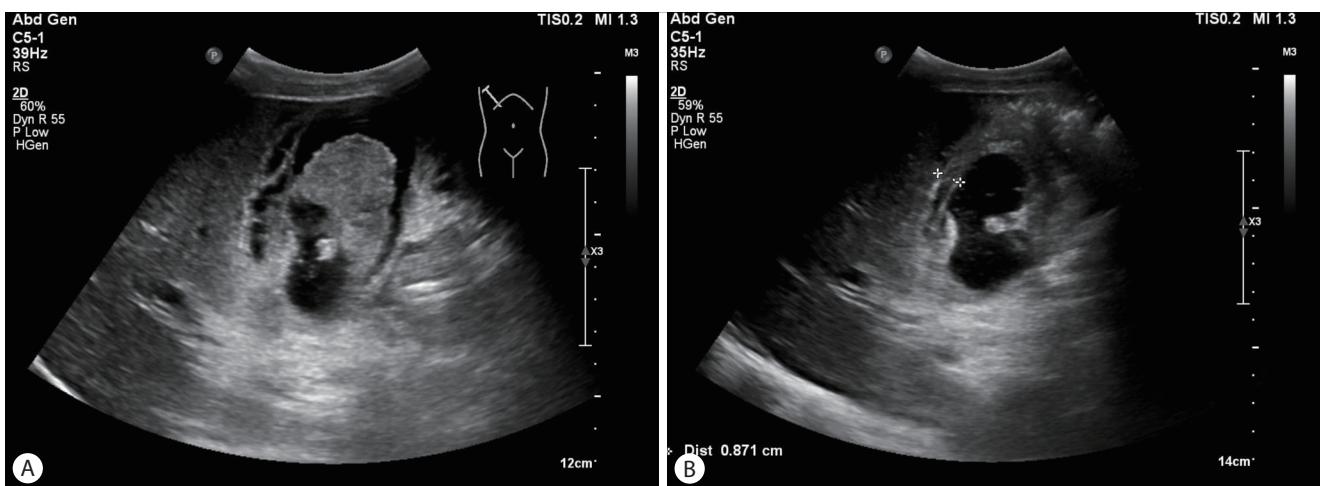


Fig. 1. (A) Ultrasound of the gallbladder showed wall irregularity and a mass with nonshadowing nonlayering intraluminal echoes. (B) Gallbladder wall thickening measured 8.7 mm.

tenderness upon physical examination. Abdominal ultrasound revealed gallbladder with a length of 9.2 cm, and exhibited irregular gallbladder wall thickening measuring 8.7 mm. There were irregular, hyperechoic mass-like lesions immobile with position changes, with distinct borders and suspended echogenic membranous structures (Fig. 1). These findings did not show any posterior acoustic shadowing. Sonographic Murphy sign was not apparent, and the diameters of the hepatic and common bile ducts were normal. The CT revealed suspicions of gallbladder and common bile duct hemorrhage and hematoma that were absent in the previous CT, along with contrast extravasation in the gallbladder in the arterial phase (Fig. 2). After diagnosing the patient with the suspicion of acalculous cholecystitis, and simultaneously considering the patient's perioperative risk of

pulmonary complications, which was assessed as high using the ARISCAT score, percutaneous transhepatic gallbladder drainage was performed on the same day as a preemptive measure. Laparoscopic cholecystectomy was performed two days later. The dissected specimen showed a lumen filled with a hematoma, hyperemic and swollen mucosa, and acute pyogenic cholecystitis, but no gallstones (Fig. 3). After decompression and surgery, LFT findings returned to normal. The patient remained stable through antibiotic therapy for the underlying infection and was discharged. The patient is currently undergoing follow-up at outpatient clinic.

DISCUSSION

HC is a rare form of cholecystitis, and most cases show gallstones

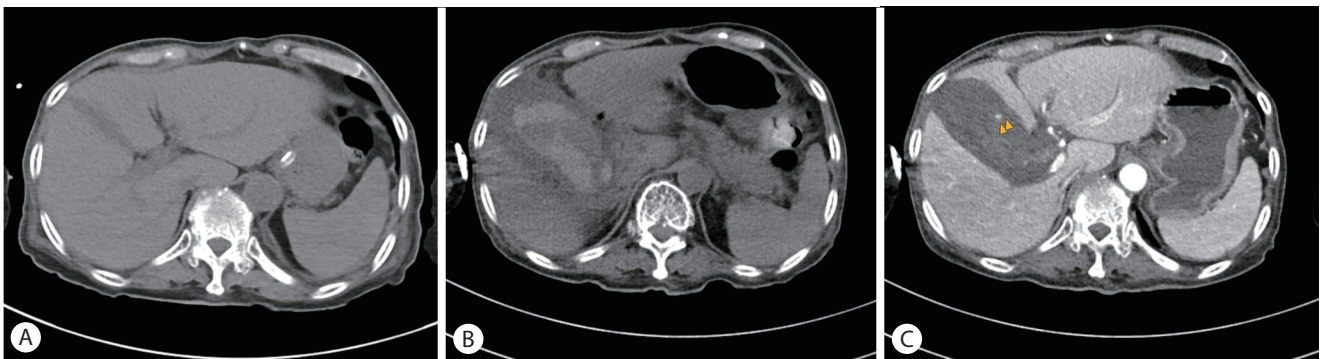


Fig. 2. (A) Non-contrast phase computed tomography (CT) in the emergency department before admission showed no abnormal gallbladder findings. (B) CT performed after ultrasound revealed increased density of sludge-like material in the gallbladder cavity and peri-gallbladder effusion in hospital day 28. (C) Suspected contrast extravasation in the gallbladder was exhibited during the arterial phase (arrowheads). After percutaneous cholecystostomy, approximately 8 mL of bloody bile was discharged, with the remainder being assessed as blood clots.

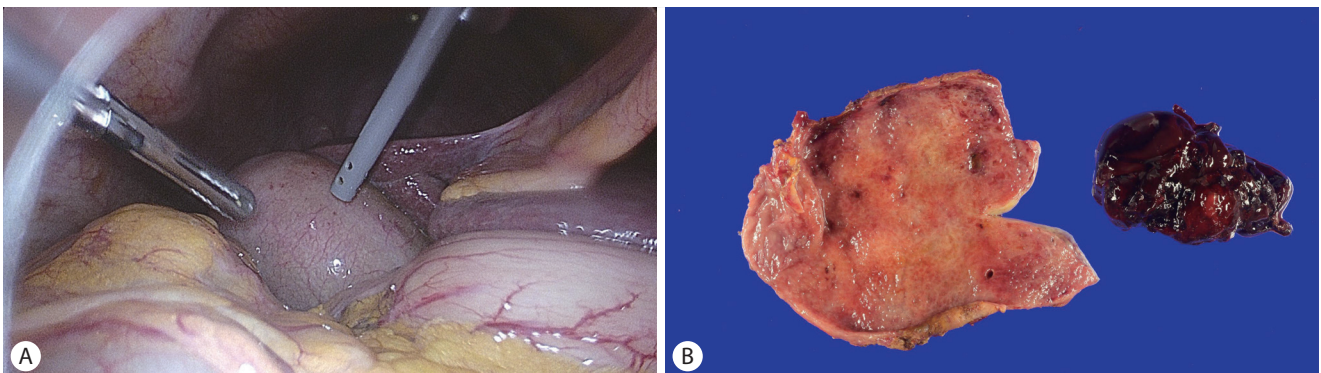


Fig. 3. (A) Findings during the laparoscopic cholecystectomy showing distended and congested gallbladder. (B) Gross appearance of the dissected gallbladder and gallbladder hematoma. The dissected specimen was measured 11.1×4.1 cm and the histopathologic examination showed findings consistent with acute suppurative cholecystitis.

with high mortality and complication rates. In a retrospective study of 6,458 patients who underwent cholecystectomy between 2000 and 2021, Hotak et al.⁷ reported that 35 patients were diagnosed with HC, showing an overall incidence of 0.55%. Among these patients, 31 out of 35 (88.6%) presented as emergency cases. Prior to surgery, 23 patients underwent abdominal ultrasound, 12 had CT scans, and three underwent magnetic resonance imaging/magnetic resonance cholangiopancreatography examinations. However, preoperative imaging dated to the diagnosis of gallbladder bleeding only in one case.⁷

Gallbladder bleeding is a rare phenomenon and is mostly associated with complications of cholecystitis, which are often triggered by the presence of gallstones. In situations where there are no gallstones, gallbladder bleeding can occur if there are risk factors such as trauma, biliary tumors, vascular disease, the use of anticoagulants or antiplatelet drugs, or biliary parasitic infections.¹

In cases of noncalculous cholecystitis without risk factors, spontaneous gallbladder bleeding is believed to occur along with mucosal injury. It is thought to be caused by inflammation of the gallbladder wall, mucosal necrosis, and subsequent damage to the blood vessels within the gallbladder wall. In some cases, aneurysmal change in cystic artery can result in bleeding into the lumen.^{5,8-10} According to a research that compiled risk factors in case reports of HC, approximately 45% of cases were found to be associated with anticoagulant therapy.⁶ The present case occurred in a patient who had not taken antiplatelet agents or anticoagulants and had no bleeding tendency or other stated risk factors; moreover, the patient was a hospitalized patient without a trauma episode linked to the development of cholecystitis.

In a study analyzing 19 cases in which HC was diagnosed histopathologically, Chinn et al.¹¹ reported the presence of gallstones in 17 cases; although the clinical manifestations, such as gastrointestinal bleeding and hemobilia did not differ between HC and non-HC, the authors observed that 74% of HC cases showed ultrasound findings such as focal gallbladder wall irregularity, intraluminal membranes, and nonshadowing nonlayering intraluminal echoes. Gremmels et al.⁹ suggested that an organized thrombus in HC must be differentiated from a polypoid intraluminal mass. While blood flow examination using Doppler can be used for this purpose, it is notable that pseudo-artery signs

identified through Doppler can also be overlapped by a vessel lying within sloughing mucosa in gangrenous cholecystitis.^{9,12} Contrast-enhanced ultrasound can be helpful in diagnosing pool-like contrast leakage in cases of HC.¹³ In noncontrast CT scans, it is possible to observe high-density intravesicular findings of blood clots. These findings can potentially be differential diagnoses for previously administered contrast agents or milk of calcium bile.^{10,12} However, these findings typically do not exhibit similar echogenicity on ultrasound as blood clots, unless there are calcium bilirubinate and cholesterol crystals within the sludge, and they may be located in gravity-dependent areas with posterior acoustic shadow. These characteristics can be used to differentiate gallbladder bleeding by comparing CT and ultrasound findings, still do not constitute a specific sign.¹⁴ In contrast-enhanced CT, the term “pseudo-vein sign,” derived from angiographic findings, refers to the appearance of extravascular leakage when blood fills the lumen of an organ. This sign can be observed in gallbladder when there is extravasation of contrast material from the blood vessel. Additionally, CT scans can also visualize pseudoaneurysms of cystic artery within the gallbladder.^{5,9,10,12} Magnetic resonance imaging findings of HC, T1-weighted image can observe high signal intensity of methemoglobin while T2-weighted image can observe hypointense clots.¹² In our case, focal gallbladder wall irregularity, intraluminal membranes, and nonshadowing nonlayering intraluminal echoes were all observed, but in Doppler signals, blood flow was not clearly detected, and extravascular leakage was confirmed on CT scan.

Bleeding in the gallbladder shows diverse courses of progression. Hematoma enlargement within the gallbladder may distend and perforate the gallbladder and cause hemoperitoneum¹⁵ and occlusion of the common bile duct may cause obstructive jaundice. In contrast, blood flowing to the small intestinal lumen may cause hematemesis or hematochezia.^{4,12} A review of 30 cases of HC from studies published from 1985 to 2018 reported that most patients (n=22, 71%) were treated with a cholecystectomy including twelve cases of open cholecystectomy, compared to three patients (10%) were treated with a percutaneous cholecystostomy drainage.⁶ It is important to have active interest in whether HC progresses from the early stages of cholecystitis with hemodynamic instability.

In general, acute acalculous cholecystitis is discovered late, as shown by more than 50% prevalence of complications such as gangrene at the time of diagnosis. It is known that the prognosis is particularly worse when the time taken to diagnosis is longer than 48 hours.^{8,10} The patient in our case also did not show typical right epigastric pain or fever, and the gallbladder inflammation was first detected during a point-of-care ultrasound performed to explore abnormal LFT findings during a hospital stay. Subsequently, the patient was immediately treated through percutaneous gallbladder drainage and follow-up cholecystectomy, which helped prevent severe complications. As shown by previous studies, HC is not diagnosed in many cases until the postoperative pathological study, while acalculous cholecystitis is relatively rare.⁷ Active suspicion and the appropriate use of point-of-care ultrasound for high-risk patients may be helpful for early diagnosis and the promotion of a better disease prognosis.

요약

출혈성 담낭염은 담낭염의 형태 중 드문 것으로, 특히 그동안 출혈성 무결석 담낭염에 대한 보고는 제한적으로 이루어져 왔다. 본 증례보고에서는 척추추간판염으로 입원 중인 환자에게서 간기능 검사의 이상 소견이 발생하여 시행한 현장초음파상에서 보인 이상 소견을 기반으로 출혈성 담낭염을 의심하였다. 환자는 문헌상 초음파상에서 출혈성 담낭염의 특징으로 제시된 담낭벽의 불규칙한 비후, 후방 음영이나 층적을 동반하지 않는 내강 안 에코의 소견 등을 만족하였으며, 이어서 촬영한 CT에서도 조영제 누출의 소견이 보여 혈액학적으로 안정된 상태에서 조기에 경피적 배액술과 후속 담낭절제술을 시행하였다. 절제된 검체에서는 혈괴와 함께 화농성 담낭염 소견이 확인되었다. 기존에 항혈소판제, 항응고제 등의 약제 복용이나 알려진 출혈 경향이 없었던 화농성 척추추간판염 환자에게서 출혈성 무결석 담낭염을 현장초음파로 합병증 발생 전에 진단하여 치료한 증례로서 보고하는 바이다.

국문 색인: 무결석담낭염; 혈액담즙증; 복강경담낭절제

Conflicts of Interest

The authors have no conflicts to disclose.

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