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Early Carcinoma of the Gallbladder Accompanied by Hemobilia: Report of a Case

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Abstract: The majority of cases of hemobilia are of traumatic or vascular origin; however, we report herein the rare case of a 51-year-old man with early carcinoma of the gallbladder accompanied by hemobilia. He had been diagnosed as having gallbladder polyps during an admission to a local hospital for treatment of cardiac failure. Preoperative evaluation in our hospital suggested that the polypoid lesion was an advanced carcinoma of the gallbladder. Laparotomy revealed that the gallbladder contained blood clots, and a cholecystectomy was performed. Histological examination confirmed the main lesion to be IIb-type early carcinoma of the gallbladder and proved that the blood clots had been misinterpreted as polyps or advanced carcinoma before the operation. The diagnostic approaches and characteristics of hemobilia in the gallbladder are discussed with a review of the literature.

Key Words: carcinoma of the gallbladder, hemobilia, gallbladder hemorrhage

Introduction

Carcinoma of the gallbladder is asymptomatic in its early phase, and establishing a clinical diagnosis is often difficult. An abnormal communication between the biliary tract and blood vessels may cause hemobilia, the etiologies of which include trauma, vascular lesions, inflammatory disorders, and neoplasms. Early carcinoma of the gallbladder could be a pathological entity of hemobilia. Various diagnostic modalities are available, and the first step toward the early detection of hemobilia is to recognize and investigate the idea of this clinical entity.

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Case Report

A 51-year-old man was referred to our hospital on July 23, 1994, for further evaluation and treatment of polypoid lesions of the gallbladder. On June 10, 1988, he had undergone an appendectomy at our hospital, at which time a complete A-V block had also been identified. A cardiac pacemaker had been implanted 12 days after his appendectomy. Following his discharge from the hospital, he had regularly been followed up at a local hospital. In 1993, at the age of 50 years, he was admitted to the local hospital for treatment of cardiac failure. During his hospital stay, a small gallbladder polyp was noted by abdominal ultrasonography (US). Following discharge, he underwent regular US examinations to observe the gallbladder polyp, and 7 months later, the polyp was found to be enlarged with a lobulated surface, indicative of a gallbladder carcinoma, whereupon he was admitted to our hospital.

He had no symptoms and a physical examination showed that he was afebrile and his vital signs were normal. The laboratory studies revealed a hemoglobin of 14.3 g/dl, a hematocrit of 41.3%, a white blood cell count of 3840/mm^3, a platelet count of 7.9×10^4/mm^3, a total bilirubin of 0.7 mg/dl, a glutamic oxaloacetic transaminase level of 27.9 IU/l, a glutamic pyruvic transaminase level of 24.8 IU/l, and a gamma-glutamyl transpeptidase level of 82.7 IU/l. The prothrombin time, partial thromboplastin time, and hepaplastin test were within normal limits. There were no abnormal values in the tumor markers such as CEA, CA 19-9, and DUPAN-2. The patient had been taking 0.25 mg/day of digoxin, and the serum level of digoxin was 2.1 ng/ml, the normal range being 0.8–2.0 ng/ml. Abdominal US revealed two echogenic intraluminal masses in the gallbladder (Fig. 1); one was a well-outlined mass 1 cm in diameter, located on the wall of the hepatic side of the neck, and the other was an oval-shaped mass with an irregular surface, 3.7 cm in length, located on the wall of

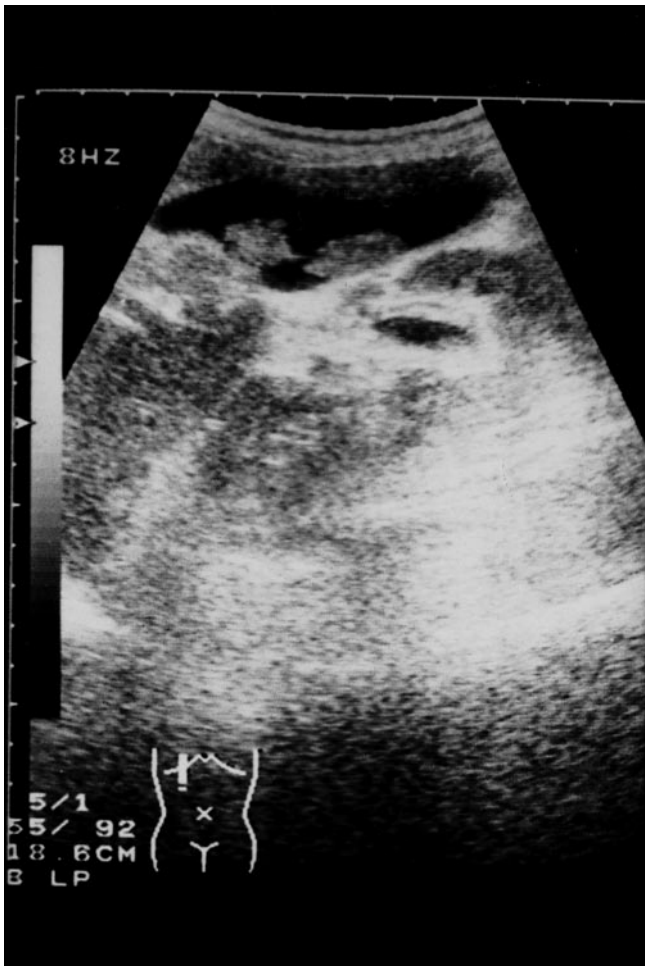

Fig. 1. Sagittal sonogram. Two echogenic masses were seen in the gallbladder; one was a well-outlined mass in the neck, 1 cm in diameter, and the other, an oval-shaped mass in the body, 3.7 cm in length. Both lesions were inhomogeneous and cast no acoustic shadows

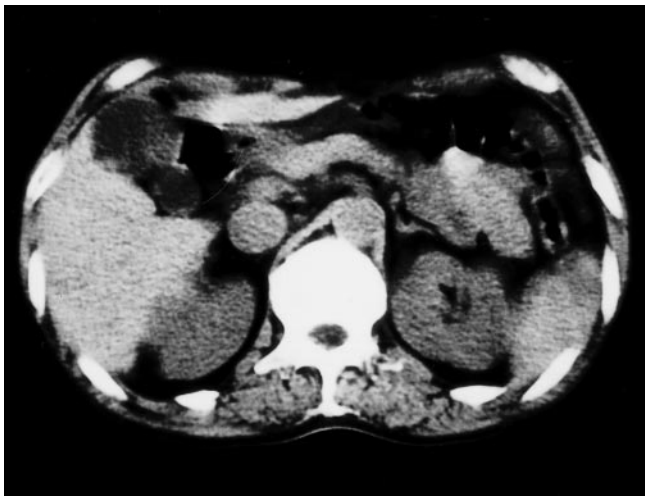

Fig. 2. Abdominal computed tomography scan revealed hyperdense masses in the gallbladder

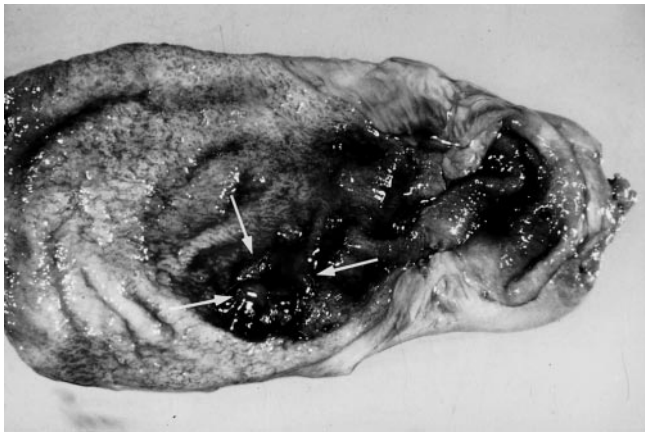

Fig. 3. IIb-type early carcinoma in the body of the gallbladder, measuring 12 mm × 12 mm (*arrows*)

the peritoneal side of the body. Both lesions were inhomogeneous and had no acoustic shadows. There was no thickening or irregularity of the wall. A plain abdominal computed tomographic (CT) scan showed spontaneously hyperdense masses in the gallbladder (Fig. 2). Endoscopic retrograde cholangiopancreatography (ERCP) was performed, which demonstrated that although the common bile duct was normal, the contrast material did not enter the gallbladder. Selected hepatic arteriography demonstrated no displacement of the right hepatic artery or cystic artery.

A laparotomy was performed under the diagnosis of "papillary tumor of the gallbladder." The liver was markedly congestive despite preoperative treatment with digoxin and diuretics. The gallbladder contained a soft palpable mass, but there was no invasive lesion on the wall. To avoid bleeding from the congestive hepatic surface, a cholecystectomy was performed first, which revealed that the lumen of the gallbladder was filled with blood clots. A small clot, 6 mm in diameter, remained on the mucosal surface after the gallbladder had been washed with saline. An immediate frozen-section examination revealed a well differentiated adenocarcinoma. The main lesion was a IIb-type early carcinoma[1] in the body of the gallbladder, measuring 12 mm × 12 mm, which was microscopically confined within the mucosa (Fig. 3). The clot involved tiny fragments, less than 6 mm in diameter, of carcinoma tissue (Fig. 4). A lymphadenectomy in the hepatoduodenal ligament was performed without further hepatectomy. Subsequent histology revealed that the tubular adenocarcinoma partially containing papillary adenocarcinoma was limited to the mucosa and Rokitansky-Aschoff sinuses with no lymph node

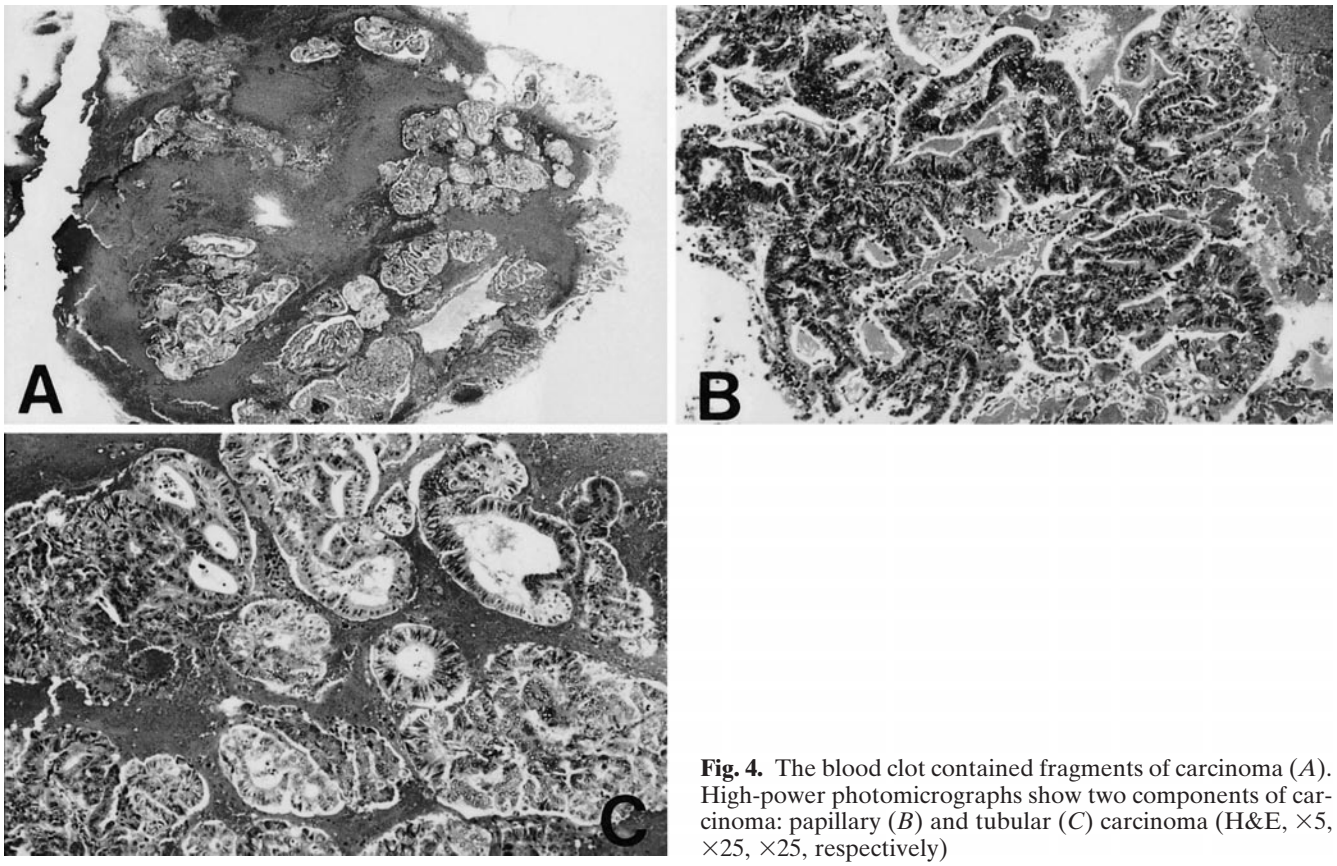

Fig. 4. The blood clot contained fragments of carcinoma (*A*). High-power photomicrographs show two components of carcinoma: papillary (*B*) and tubular (*C*) carcinoma (H&E, ×5, ×25, ×25, respectively)

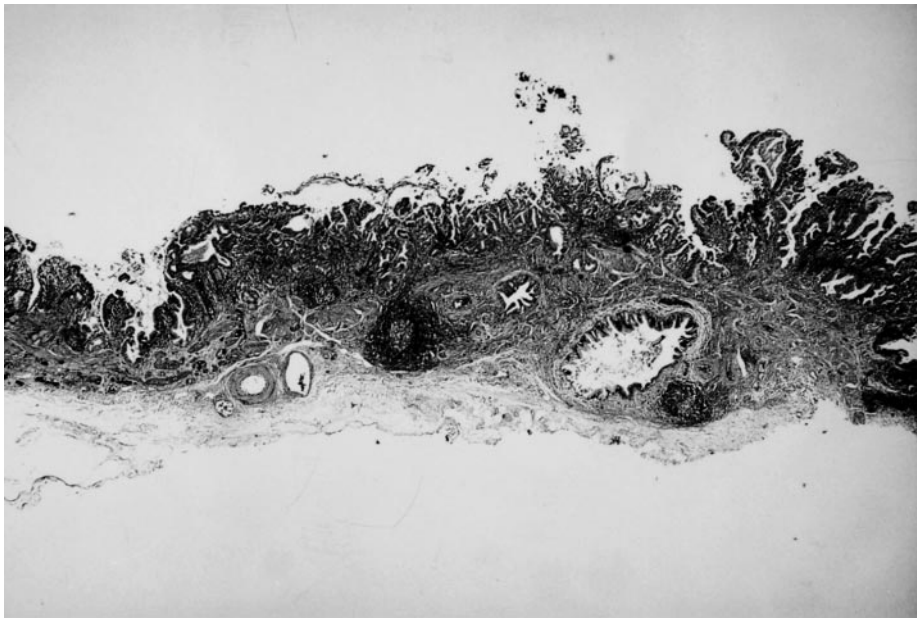

Fig. 5. IIb-type early carcinoma in the body of the gallbladder with invasion limited to mucosa and Rokitansky-Aschoff sinuses (H&E, ×5)

metastasis (Fig. 5). The hemobilia was suspected to have originated from the surface of the carcinoma. The patient had an uneventful postoperative recovery and has remained well since discharge from the hospital.

Discussion

Since Sandblom[2] coined the term "hemobilia" in 1948 to describe hemorrhage into the biliary tract, a large number of cases of hemobilia have been reported. The classic clinical triad of hemobilia consists of gastrointestinal bleeding (90%), biliary colicky pain (70%), and jaundice (60%).[3] The bleeding may originate from within the liver, the vessels in the hepatoduodenal ligament, the common bile duct, the gallbladder, or the pancreas. Hemobilia accounts for 2%–5% of all gastrointestinal hemorrhages, and the mortality rate approached 30% at the time of Sandblom's report.[4] Because 5%–10% of patients with gastrointestinal hemorrhage remain undiagnosed, some cases of hemobilia have been classified as "bleeding of unknown origin".[5] The causes of hemobilia include trauma (39%), surgery (17%), calculi (15%), inflammation (13%), vascular disorders (11%), and neoplasms (5%)[6]. In neoplasm-related hemobilia, the gallbladder tumor shares a 24% incidence rate;[6] however, hemobilia following carcinoma of the gallbladder is a rare phenomenon.

For the diagnosis of hemobilia originating from the gallbladder, various diagnostic techniques are available. Abdominal US may reveal intraluminal, fixed, and nondependent hyperechoic lesions without acoustic shadows; however, stones are movable and dependent and cast acoustic shadows. Marchal[7] analyzed the sonographic aspect of hemobilia in the gallbladder and reported that the hyperechoic content of the gallbladder was seen only after a recent hemorrhage, and that rapid change into a less reflective, non-shadowing intraluminal mass should be regarded as diagnostic of hemobilia. Abdominal CT scan may reveal an intraluminal mass in the gallbladder and common bile duct; however, ERCP will verify bleeding from the papilla of Vater, demonstrate blood clots in the gallbladder and common bile duct, and often contribute to identification of the etiology.[8] Angiography may identify the vessel lesion from which the bleeding originates, and make visible the contrast medium passage into the biliary tract.[9]

The definition of early carcinoma of the gallbladder, according to the General Rules for Surgical and Pathological Studies on Cancer of Biliary Tract,[1] is as follows: "Its invasion is histologically confined within mucosa and muscularis propria, regardless of lymph node metastasis." A review of 148 cases of hemobilia collected by Senda[10] from the Japanese literature before 1984 revealed only four patients with carcinoma of the gallbladder from which bleeding originated. In our literature review, from the end of Senda's series up to May 1996, a further ten patients who developed hemobilia from carcinoma of the gallbladder, including three with early carcinoma, were reported.[11–13] In these three patients, intraluminal masses were detected by abdominal US, and the histological diagnosis was well-differentiated adenocarcinoma. Two of them had type I early carcinoma[1] with invasion limited to the mucosa and muscularis propria, respectively, while the third had type IIa[1] with invasion deep in the muscularis propria. Fragments of carcinoma tissue were found in the hemobilia in all three cases.

The patient reported here is therefore only the fourth case of hemobilia from early carcinoma of the gallbladder to be reported in Japan. Aside from the fragments of carcinoma tissue found in the blood clot, postoperative histological examination disclosed no definite origin of the bleeding, such as necrosis, ulceration, or submucosal hemorrhage of the gallbladder wall. It is suggested that after small pieces of tissue fell off of the surface of the carcinoma, an insignificant amount of blood leaked into and formed blood clots in the gallbladder over the course of 1 year. From his experiment, Sandblom[14] described the difference in the fate of clots in the biliary tract. When clots are continually washed in bile, they disappear quickly, but they stay intact when they are protected from the bile stream. In our patient, clots were formed and remained only in the gallbladder away from the bile stream. This may account for their having been misidentified as polyps or advanced carcinoma preoperatively.

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