

[CASE REPORT]

Liver Cyst Infection after Colon Endoscopic Mucosal Resection in a Patient with Autosomal Dominant Polycystic Kidney Disease on Maintenance Hemodialysis

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Abstract:

A 60-year-old Japanese man with autosomal dominant polycystic kidney disease (ADPKD) on maintenance hemodialysis underwent colonoscopy and endoscopic mucosal resection (EMR). He was hospitalized after 4 days of fever that began the day following colonoscopy. We detected *Klebsiella pneumoniae* in a blood culture and a ring-shaped integration in the liver cyst by gallium scintigraphy. He recovered with antibiotics and percutaneous drainage. The patient was believed to have contracted the liver cyst infection via an injured colonic mucosa and portal vein. Thus, if a patient exhibits fever after a colon EMR, for patients with ADPKD, then a liver cyst infection should also be considered in the differential diagnosis.

Key words: autosomal dominant polycystic kidney disease, liver cyst infection, endoscopic mucosal resection, *Klebsiella pneumoniae*, colonoscopy, polypectomy

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Introduction

Cyst infection is a serious complication in autosomal dominant polycystic kidney disease (ADPKD), with a reported incidence of 0.01 episode per patient per year (1). Colonoscopy and endoscopic mucosal resection (EMR) have been established as useful, minimally invasive, and safe treatments for ADPKD; however, complications with bacteremia (2-4) and a small number of liver abscesses arising from these procedures have been reported (5-11). We herein describe a patient with ADPKD on hemodialysis who experienced liver cyst infection after undergoing colon EMR.

Case Report

A 60-year-old Japanese man on maintenance hemodialysis due to ADPKD was admitted to our hospital for an evaluation of fatigue, fever, and inflammation. He had a 30-year history of hypertension under medication. At 51 years of

age, he was admitted with multiple cysts of the liver and kidney and was diagnosed with ADPKD; he began hemodialysis at 53 years of age.

He underwent colonoscopy in August 2014 because of a positive fecal occult blood test, revealing four colonic polyps: 8 mm Isp in the cecum, 10 mm Ip at the sigmoid-descending junction, and 10 mm Isp and 20 mm Isp in the sigmoid colon. These were treated with polypectomy or EMR. The patient developed a fever of 38.5°C on the day after the procedure and on the fourth day was admitted to our hospital for an evaluation of the fever and high-grade inflammation. He had no prior history of cyst infection or other similar diseases.

On admission, the patient suffered from fatigue and a temperature of 38.4°C, and his blood pressure was 81/57 mmHg. He had no other symptoms, including abdominal pain, tenderness, and liver percussion pain. The laboratory results were as follows: his white blood cell count of 3,500/ μ L was in the normal range, but neutrophils increased to 94.8%, and C-reactive protein was markedly elevated to

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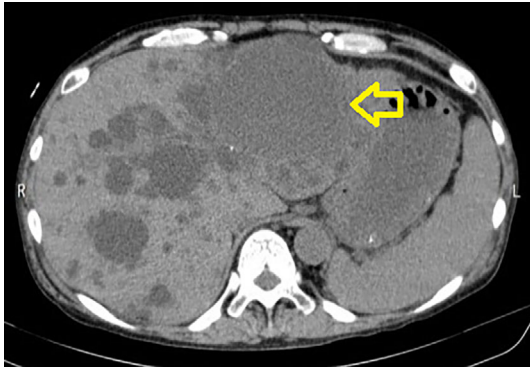


Figure 1. Abdominal computed tomography taken on the day of hospitalization revealed numerous cysts in the liver; however, there was no evidence to indicate a cyst infection arrow: infected cyst.

30.64 mg/dL. Total bilirubin, aspartate aminotransferase, alanine aminotransferase, alkaline phosphatase, and γ -glutamyl transpeptidase were in the normal range. Abdominal computed tomography (CT) revealed numerous cysts in the kidneys and liver, but there was no evidence to rule out a cyst infection, with neither thickening of the cyst walls nor elevated concentration of the internal fluid (Fig. 1), or other focus of infection, including the lung, kidney, colon, or other thoracoabdominal or pelvic organs.

The clinical course is shown in Fig. 2. Septic shock was suspected and the patient was treated with sulbactam-cefoperazone even though it was not possible to identify any obvious focus of infection. However, the inflammation did not improve. As two sets of blood cultures were positive for *Klebsiella pneumoniae*, on hospital day 7 the antibiotic therapy was changed to levofloxacin (LVFX) for a more focused antibiotic spectrum. Gallium scintigraphy was performed on day 13, thus revealing an abnormal annular accumulation at the upper abdominal midline; in conjunction with CT results, the patient was diagnosed with liver cyst infection (Fig. 3).

Percutaneous liver cyst drainage was performed on hospital day 17, thereby releasing a brown purulent fluid that contained a high concentration of neutrophils. A culture of the liquid content was negative and no malignancy was observed. After drainage, the inflammation gradually subsided and the drainage tube was removed on day 33 (Fig. 4). The patient was discharged on day 41 and LVFX was discontinued on day 51.

As a moderately differentiated tubular adenocarcinoma with vein invasion was identified from the 20-mm polyp resected by EMR from the sigmoid colon, a laparoscopic high anterior resection was performed on day 65.

Discussion

We herein report a case of hepatic cyst infection after colonoscopic polypectomy in a patient with ADPKD on maintenance hemodialysis. Liver cysts are present in ap-

proximately 80% of ADPKD patients, representing the most common extrarenal manifestation of ADPKD (12). A hepatic pyocyst is a potentially life-threatening complication of ADPKD.

The etiology of this case was thought to be transient bacteremia arising from the EMR which led, via the portal vein, to an infection of the liver cyst, with renal failure and ADPKD as possible exacerbating factors. Colon EMR can result in the development of liver infection by bacteremia. The etiology of liver abscesses has previously been categorized according to the infection route: 1) through the bile duct, 2) through the portal vein, 3) by direct extension, 4) from blunt or penetrating trauma, 5) through the hepatic artery, or 6) of obscure origin, cryptogenic (13). With regard to the portal vein infection route, the key process is considered to be a mucosal defect presenting within digestive tract lesions or a compromised mucosal barrier that allows a route for bacteria invasion into the portal system with subsequent hematogenous spread to the liver (14). A recent study revealed that 2-4% of asymptomatic bacteremia arose during colonoscopy (2-4). In a study of 37 patients with pyogenic liver abscesses, 24% were diagnosed with a colonic cause based on the recognition of lesions with colonic mucosa defects, such as cancer (15). In the present case, the liver cyst infection was thought to be due to bacteria entering the blood from mucous membrane defects caused by the EMR and then passing through the superior mesenteric vein and the portal vein to reach the liver.

Although perforation and peritonitis are common infectious complications after EMR, we have found no previous case reports of liver cyst infection. Reports of only six liver abscess cases that developed after colonoscopy have been found (5-11). In these, the time from colonoscopy to fever was 12 hours to 5 days. Two of the three cases of polyps that underwent pathological examination showed a malignant tumor. All the liver abscesses occurred in the right lobe and they improved after the administration of antibiotics and drainage. Our case showed a similar time from colonoscopy to fever as the previous reports and was similarly improved by antibiotics and drainage; however, the patient in this case had a cyst infection rather than an abscess. Furthermore, he also differed from the previous reports due to the fact that he had ADPKD and was also on hemodialysis.

ADPKD and end-stage renal disease can be contributory factors to liver infection after colon EMR. Previous studies of rat uremia have revealed the disintegration of colonic (16), gastric, and small intestinal (17) epithelial tight junctions, suggesting that renal failure may be related to a decrease in the intestinal barrier function. Furthermore, a study examining blood bacterial DNA during dialysis reported that bacteremia from the digestive tract, related to chronic inflammation, occurred in 20% of all asymptomatic hemodialysis patients (18, 19). Bacteria from the intestinal tract (such as *Escherichia coli*) are frequently reported as a cause of liver cyst infection in ADPKD patients (1, 20). In this case, renal failure was considered to be a precipitating

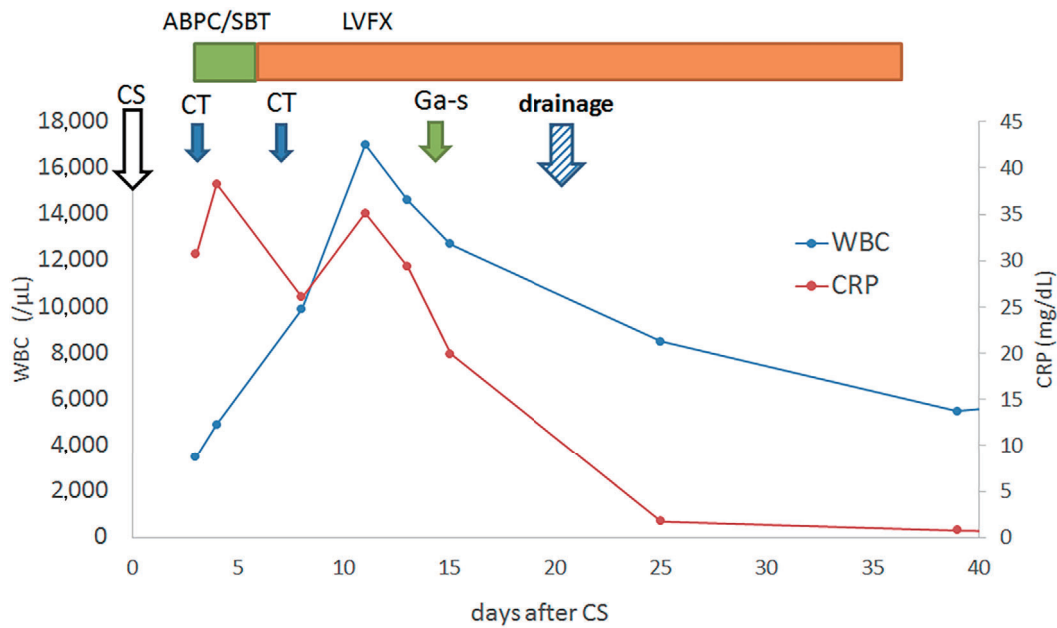


Figure 2. Clinical course. ABPC/SBT: sulbactam/ampicillin, CRP: C-reactive protein, CS: colonoscopy, CT: computerized tomography, Ga-s: gallium scintigraphy, LVFX: levofloxacin, WBC: white blood cell count



Figure 3. Gallium scintigraphy showing annular accumulation in the liver cyst.



Figure 4. Abdominal computed tomography after treatment of the liver cyst infection.

factor for translocation and the subsequent bacteremia and liver cyst infection.

It is possible that the liver cyst infection may have developed from a cause other than EMR. However, we believe that this infection derived from colon EMR for the following reasons: First, the time period from colonoscopy to the development of fever is an important factor. Even though the patient had never previously experienced a cyst infection, he was admitted because of fever that had developed the day after undergoing endoscopic colonoscopy and EMR.

Because the incidence of cyst infections in patients with ADPKD was 0.01 episode per patient per year (1), the possibility that some coincidence occurred by chance is therefore difficult to assume. Second, the causative bacteria, *Klebsiella pneumoniae*, can colonize the gastrointestinal tract of humans, and liver infection caused by this bacterium may be a result of translocation from the intestinal tract (21). Third, other sites that may be a source of infectious diseases were not considered. For the onset of a liver infection, the abovementioned pathway is necessary for the invasion of the pathogenic bacteria; however, the patient had no abnormality in the bile duct or organs around the liver; no trauma other than to the colon mucosa, which was damaged because of EMR; and no infection causing bacteremia, which was revealed by physical findings and whole-body CT. Therefore, we believe that liver cyst infection in this case had been caused by colon EMR.

It is uncertain whether colonoscopy itself or EMR caused

this cyst infection. In the review of 12 studies investigating the frequency of colonoscopy and bacteremia, bacteremia was found in 2.2% of the colonoscopy cases; however, there was no correlation of bacteremia with biopsy, polypectomy, or the length of the instrument inserted (4). In addition, one case report described a liver abscess that had developed due to screening-only colonoscopy without any invasive procedures, such as biopsy or EMR (11), but most of the other reported cases described procedures which may cause an injury to the colon mucosa, such as EMR. In the present case, we believe that EMR is a highly possible cause because of the degree of invasion, namely, the magnitude of the risk of translocation. In conclusion, we herein described a case of hepatic cyst infection in a patient with ADPKD on maintenance hemodialysis after colonoscopic EMR. Although most bacteremia after colonoscopic polypectomy is transient, it can sometimes cause severe infectious complications. Thus, if a patient is admitted with fever after colonoscopy, then systemic infections should be considered; furthermore, liver cyst infection should also be considered for ADPKD patients.

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

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