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CASE REPORT

Severe refractory infection due to renocolic fistula in a patient with a giant kidney and ADPKD undergoing long-term hemodialysis

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Abstract
Renocolic fistula is rare. Renal cyst infection is a serious complication in patients with autosomal dominant polycystic kidney disease (ADPKD). We present a case of refractory renal cyst infection due to renocolic fistula in a patient with ADPKD. A 65-year-old man with ADPKD on hemodialysis visited our hospital with complaints of fever and left abdominal pain. We diagnosed renal cyst infection with abdominal computed tomography scans. After hospitalization, gas shadow was observed in the left renal cyst. Percutaneous puncture of the cyst was performed. Because contrast medium into the left renal cyst through nephrostomy was flowing into the descending colon, renocolic fistula was diagnosed. The patient underwent nephrectomy combined with partial descending colonic resection and splenectomy, but he died. Renocolic fistula is probably hidden in some refractory renal cyst infection cases. This case report aims to create awareness of renocolic fistula, so that early diagnosis and intervention can salvage such patients.

Keywords Autosomal dominant polycystic kidney disease · Gas in cyst · Hemodialysis · Renal cyst infection.

Introduction

Renal cyst infection is one of the most serious complications in patients with autosomal dominant polycystic kidney disease (ADPKD). A total of 30–50% of patients with ADPKD experience some form of kidney infection during their lifetime [1–3]. Development of a fistula between the colon and kidney, called renocolic fistula, is rare. We present a case of refractory renal cyst infection due to renocolic fistula in a patient with ADPKD on long-term hemodialysis (HD).

Case report

A 65-year-old man with end-stage renal disease due to ADPKD visited our hospital with complaints of fever and left abdominal pain. He had been treated with HD three times per week for 34 years. He had a past history of right nephrectomy because of renal cancer in 1998 and permanent pacemaker implantation because of Complete atrioventricular block in 2010. We diagnosed left renal cyst infection with abdominal computed tomography (CT) scans (Fig. 1). Initially, we treated him with levofloxacin. His fever persisted and he gradually weakened. We added metronidazole to levofloxacin, but it was not effective, and he was hospitalized. Laboratory test results on admission were as follows: white blood cell count of 4800 /μL, hemoglobin of 10.1 g/dL, and C-reactive protein level of 12.93 mg/dL. After hospitalization, gas was observed in the left renal cyst in abdominal CT on day 11 (Fig. 2). Because we considered that we could not treat him with only antibiotic therapies, percutaneous cyst drainage and cystic imaging were performed. The presence of a renocolic fistula was detected, because contrast medium into the left renal cyst through nephrostomy was flowing into the descending colon (Fig. 3). At first, serous fluid was discharged, and the fluid gradually became like feces. Culture

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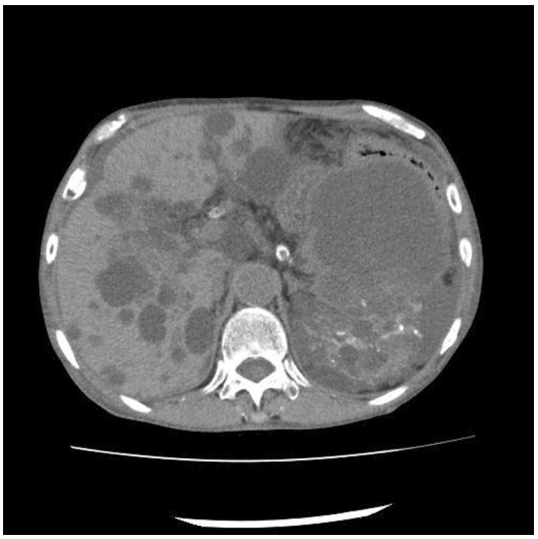

Fig. 1 Initial abdominal CT scans. One of the renal cysts in the left kidney is shown as a slightly high density area compared with the other cysts

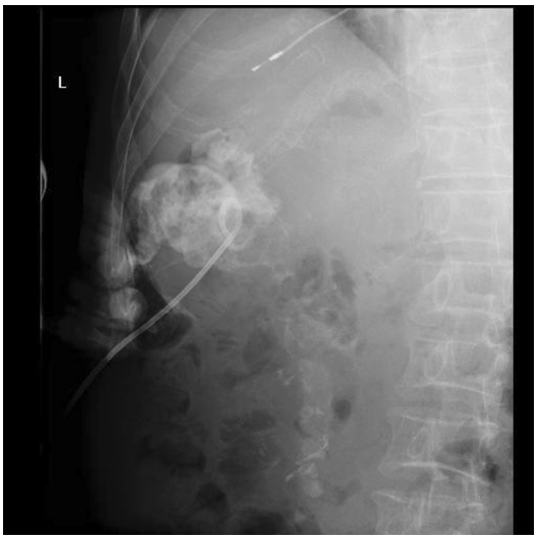

Fig. 3 Injection of contrast medium into the cyst through a pig-tail catheter shows accumulation of contrast medium in the cyst and opacity in the descending colon, indicating formation of a fistula

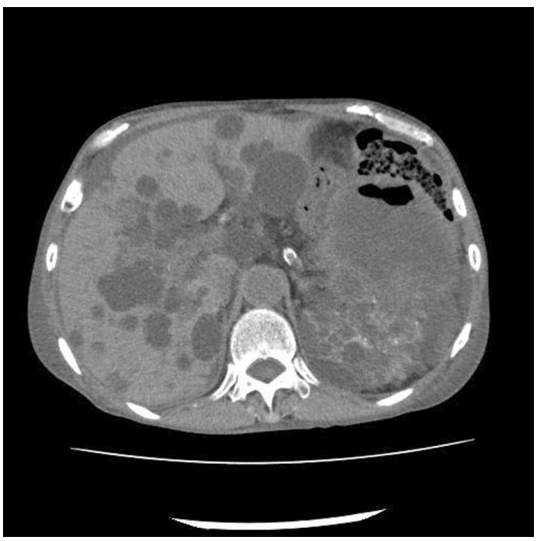

Fig. 2 Abdominal CT scans. Gas shadow was observed in the infection renal cyst

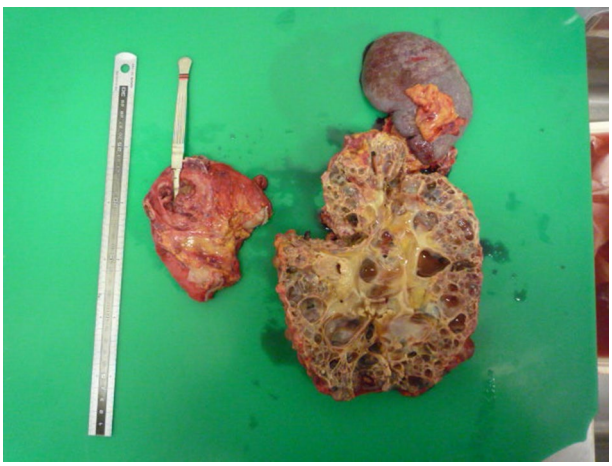

Fig. 4 Left nephrectomy, partial resected descending colon, and splenectomy specimen demonstrating renocolic fistula

of the serous fluid showed α-*Streptococcus, Bifidobacterium* species, and *Bacteroides thetaiotaomicron*. He showed the decrease of the coagulation function after hospitalization. We thought that he was a state of sepsis and DIC due to the infection. However, we believed that we could not improve the patient's condition with the current conservative treatment. He underwent nephrectomy combined with partial descending colonic resection and splenectomy on day 30 (Fig. 4). The surgeon suggested construction of colonostomy, but anastomoses were undergone for his preoperative hope. The operation was favorable halfway, but bleeding easily due to the DIC. He became the shock state during the operation many times. After the operation, his condition was not improved. Transfusions and catecholamines could not make his shock state better. He died on day 42.

Discussion

Renocolic fistula can be a cause of refractory renal cyst infection. Only 100–130 cases on renocolic fistula have been reported until recently [4], and 52 cases have been reported in Japan. However, renocolic fistula in patients with ADPKD is rare and has been found only in four patients, including our patient (Table. 1). The mechanism of renocolic fistula

Table 1 Case reports of renocolic fistula in ADPKD

Author	Age	Sex	Means of diagnosis	Gas shadow in cyst	Therapy	Time to treatment	Outcome	HD or non-HD
Loftus IM et al. [9]	48	M	Surgery	+	Surgery	6 weeks	Survive	Non-HD
Ito H et al. [6]	60	F	Barium enema	–	Surgery	22 days	Survive	HD
Ishikawa E et al. [7]	51	M	Barium enema	+	Surgery	2 weeks	Survive	Non-HD

remains unclear. A persistent inflammatory process due to renal stones, tuberculosis, and renal abscess was originally thought to occur, and then, inflammation to the colon results in a renocolic fistula [5]. However, Lee et al. reported that local abscess formation in the intestinal tract could be involved with formation of renocolic fistula [4]. Therefore, they presumed that there might be a problem on the colon side, such as diverticulum other than chronic inflammation in the renal parenchyma. Although no diverticulitis was observed in our case, we speculate that diverticula were also in the colon, where the fistula had formed, because many diverticula were observed in the ascending colon. Abdominal CT showed that the kidney widely contacted with the colon, and thus, this condition easily caused inflammation between the kidney and colon. Because renal cysts in patients with ADPKD develop tissue thinning and have a lower barrier function against bacterial infection, inflammation in renal cysts is easily prolonged. Furthermore, because patients with ADPKD have the complication of diverticula of the colon, these patients are considered to have a high risk of developing renocolic fistula.

Percutaneous drainage and cystic imaging are effective for diagnosing renal cyst infection caused by renocolic fistula. Ito et al. made the diagnosis of renocolic fistula with a barium enema examination [6]. Ishikawa et al. also made a diagnosis of hemorrhage in an infected cyst by contrast-enhanced ultrasonography, but the diagnosis of renocolic fistula was determined by an enteral barium test [7]. Renocolic fistula other than renal cyst infection is diagnosed with contrast radiography from either the kidney or intestinal tract. This diagnostic method could also be effective in the case of renal cyst infection. The present case was diagnosed by contrast radiography from a renal cyst. However, definite diagnosis may require contrast radiography from the kidney or intestinal tract. Percutaneous drainage against renal cyst infection can be effective in treatment [8]. Because a contrast agent might not appropriately reach a fistula from the intestinal tract, cystic imaging from renal cysts is desirable as a reliable diagnostic method including treatment. However, because percutaneous drainage may be difficult depending on the case, contrast radiography from the intestinal tract should be considered. There have been some reports of gas within infected cysts besides the present case [6, 9]. This is considered as a specific finding for refractory renal cyst infection caused by renocolic fistula. The reason why the color of drainage fluid changes from a slightly bloody color to watery stool-like color (or yellow ocher) could be because of intestinal tract contents flowing into the renal cyst owing to decompression of the infected cyst by drainage. When enteric bacteria sustainably flow into a naturally-aseptic kidney from the colon, it develops refractory infection and eventually leads to worsening of the general condition. The mainstay of treatment for renocolic fistula is nephrectomy combined with partial colonic resection. If renocolic fistula is small, it may be treated conservatively. Percutaneous drainage and cystic imaging were effective for a definitive diagnosis in this case. However, in a case with cyst infection due to renocolic fistula, medical treatment was ineffective, because the infected renal cyst was filled with stool. Therefore, nephrectomy should have been performed as soon as possible after confirmation.

In summary, we report a case of refractory renal cyst infection due to renocolic fistula in a patient with ADPKD. Renocolic fistula can be an uncommon cause of refractory renal cyst infection. Despite percutaneous drainage and nephrectomy, as well as partial colonic resection, our patient died. Clinicians should be aware of the possibility of renocolic fistula in case of refractory infection in patients with ADPKD.

Compliance with ethical standards

Human and animal rights This article does not contain any studies with human participants or animals performed by any of the authors.

Conflict of interest Takashi Shigematsu received a lecture fee. However, this report was not affected by receiving this fee. None of the other authors has a conflict of interests.

Informed consent Informed consent was obtained from the patient and his family included in this article.

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