Case Report
Bilateral Spontaneous Perirenal Hemorrhage in an Acquired Cystic Kidney Disease Hemodialysis Patient

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Acquired cystic kidney disease (ACKD) is a late stage complication of chronic kidney disease. Cysts tend to grow with time on dialysis and could lead to malignant transformation and intra- or perirenal hemorrhage as a rare complication of ACKD. Here we describe one case of bilateral spontaneous perirenal hemorrhage of ACKD in a 44-year-old man, on hemodialysis for 15 years. One was due to cyst rupture, and the other was due to aneurism rupture, both were controlled with transcatheter arterial embolization. In renal arteriography at the second rupture, we demonstrated extravasation from an aneurysm being present at the periphery of right renal artery. Several spontaneous perirenal hemorrhage cases were reported but its clinical information is limited, moreover, bilateral cases were extremely rare. Furthermore, to our knowledge, this is the first report of spontaneous perirenal hemorrhage caused by intraparenchymal renal artery aneurysm rupture in ACKD patients. We report this case because of its rarity and significance with respect to the complication of dialysis patients, review reported bilateral cases, and discuss some clinical characteristics.

1. Introduction
Acquired cystic kidney disease (ACKD) is a well-known late stage complication of chronic kidney disease. Cysts tend to grow with time on dialysis and could lead to malignant transformation, and intra- or perirenal hemorrhage is a rare complication of ACKD. Here we describe one case of bilateral spontaneous perirenal hemorrhage of ACKD in a 44-year-old man, on hemodialysis for 15 years. One was due to cyst rupture, and the other was due to aneurism rupture, both were controlled with transcatheter arterial embolization. In renal arteriography at the second rupture, we demonstrated extravasation from an aneurysm being present at the periphery of right renal artery. Several spontaneous perirenal hemorrhage cases were reported but its clinical information is limited, moreover, bilateral cases were extremely rare. Furthermore, to our knowledge, this is the first report of spontaneous perirenal hemorrhage caused by intraparenchymal renal artery aneurysm rupture in ACKD patients. We report this case because of its rarity and significance with respect to the complication of dialysis patients, review reported bilateral cases, and discuss some clinical characteristics.

2. Case Report
A 44-year-old man with ESRD caused by hypertensive nephrosclerosis on hemodialysis for 15 years was admitted to our hospital for sudden onset of severe left flank pain and nausea. The pain had started suddenly about 2 hours before and worsened rapidly to its peak such that he had never experienced before.

His past medical history included hyperthyroidism, bilateral subdural hematoma, and coronary heart disease. He got a coronary artery bypass grafting (CABG) 11 months before his admission. Since then, he took aspirin and warfarin daily. On admission, his blood pressure and pulse rate were 102/65 mmHg and 86 bpm, and he was symptomless except for mildly distended abdomen with severe tenderness over his left flank to back. Laboratory findings showed a low serum hemoglobin concentration 10.0 g/dL. In addition, PT-INR was slightly prolonged to 3.50. Computed tomography (CT) showed a huge hematoma extending from left kidney to perirenal and left retroperitoneal space. The scan also showed multiple cysts in both kidneys (Figure 1(a)).

Although, fluid resuscitation as well as prothrombin complex concentrates (PCC) for reversal of anticoagulation was initiated, his blood pressure declined to shock. In renal arteriography, we saw very mild extravasation at the periphery of left renal artery. We emergently performed transcatheter arterial embolization (TAE). Shortly after TAE, he recovered completely.
Seven months later, he was again admitted to the hospital because of fatigue and weakness. His blood pressure had been well controlled after discharge. On admission, no fever, vomiting, or other symptoms were present. His blood pressure and pulse rate were 92/60 mmHg, 102 bpm. His abdomen was mildly distended and mild tender to palpation in the right middle and lower quadrants. Laboratory findings showed a severe low serum hemoglobin concentration 5.3 g/dL, prolonged PT-INR 7.05, under anticoagulation therapy with warfarin. The CT scan showed a large cyst and a huge right perirenal hematoma (Figure 1(b)). The cyst, which was not found at the previous episode, showed fluid retention with density of blood. This time, in renal arteriography, we demonstrated aneurismal bleeding at the periphery of the right renal artery (Figure 1(c)). We again emergently performed TAE. After TAE, retroperitoneal hemorrhage remitted and he recovered completely in almost the same time course as the previous episode.

At outpatient review after 3 months of his discharge, the CT scan showed the contraction and absorption of retroperitoneal hemorrhage. We could not demonstrate any other cause of perirenal hemorrhage including renal cell carcinoma. He continues hemodialysis without any bleeding sign or symptom.

3. Discussion

Spontaneous perirenal hemorrhage has been reported infrequently in the literature, where dialysis patients are often excluded and most frequent etiologies are renal cell carcinoma, angiomyolipoma, and vascular disease. Among dialysis patients, ACKD has been recognized to be the most frequent cause. In the past, there have been only 4 documented cases of spontaneous bilateral perirenal hemorrhage in ACKD dialysis patients [1–4] (Table 1). All the cases were men, etiologies of ESRD were hypertensive nephrosclerosis in 4 of 5 cases (80%), and the causes of bleeding were due mainly to the rupture of renal cyst.

Interestingly, this is the first report of perirenal hemorrhage caused by intraparenchymal renal artery aneurysm rupture in ACKD patients. The large cyst including fluid with density of blood, which was not found at the previous episode, was likely to be formed by the intracystic extravasation from aneurysm. The incidence of renal arterial rupture...
Table 1: Spontaneous bilateral perirenal hemorrhage in ACKD dialysis patients.

| First Author [Ref] | Case 1 | Case 2 | Case 3 | Case 4 | Our case |
|-------------------|--------|--------|--------|--------|----------|
| Minar et al. [1]  | Carlson et al. [2] | Borras et al. [3] | Ku et al. [4] |        |          |
| Age, sex          | 35/M   | 48/M   | 37/M   | 46/M   | 44/M     |
| Race              | N.D.   | N.D.   | Black  | N.D.   | Yellow   |
| HD/PD             | HD     | HD     | PD (+5 years: HD) | HD     | HD       |
| Duration of dialysis | 5 years | N.D. | 5.5 years | 8 years | 15 years |
| Primary disease of ESRD | CGN | Hypertensive nephrosclerosis | Hypertensive nephrosclerosis | Hypertensive nephrosclerosis | Hypertensive nephrosclerosis |
| Renal Biopsy      | N.D.   | N.D.   | Yes    | N.D.   | No       |
| Past history      | N.D.   | CLL    | CH-B   | N.D.   | CABG, Hypothyroidism |
| Time interval between bilateral perirenal hemorrhage | 2 months | 4 months | 21 months | 1 month | 6 months |
| Etiology          |        |        |        |        |          |
| 1st rupture       | N.D.   | Cyst rupture | Cyst rupture | **N.D. | Cyst rupture |
| 2nd rupture       | N.D.   | Cyst rupture | Cyst rupture | **N.D. | Cyst rupture |
| Hemoperitoneum    | N.D.   | No     | Yes    | No     | No       |
| Antiplatelet agent | N.D. | N.D. | N.D. | N.D. | Aspirin, Cilostazol |
| Anticoagulant agent | N.D. | N.D. | N.D. | N.D. | Warfarin |
| Therapy           |        |        |        |        |          |
| 1st rupture       | N.D.   | ***Emergency surgery | Nephrectomy | Nephrectomy | TAE |
| 2nd rupture       | N.D.   | TAE + Nephrectomy | Nephrectomy | TAE | TAE |
| Outcome           | N.D.   | Alive  | Alive  | Alive  | Alive    |

N.D.: no data, HD: hemodialysis, PD: peritoneal dialysis, ESRD: end-stage renal disease, CGN: chronic glomerulonephritis, ACKD: acquired cystic kidney disease, CLL: chronic lymphocytic leukemia, CH-B: chronic hepatitis B, CABG: coronary artery bypass grafting, TAE: transcatheter arterial embolization.

*Article in Germany, abstract only.
*He was first treated with HD for 5 years.
**Intrakidney contrast extravasation suggestive of active bleeding was found.
***Disrupted fragments of the kidney were removed, and the renal pedicle was controlled with clamps suture ligated.

Conflict of Interests

The authors declare that they have no conflict of interests.
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