Dialysis-induced Subdural Hematoma in an Arachnoid Cyst Associated with Autosomal Dominant Polycystic Kidney Disease

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Abstract

Arachnoid cyst (AC) is a neurological complication of autosomal dominant polycystic kidney disease (ADPKD). Although an AC can increase the risk of a subdural hematoma, the clinical presentation of bleeding into an AC associated with ADPKD is not well known. We herein report the case of a 59-year-old woman in whom the initiation of hemodialysis for renal failure led to AC bleeding. A change of anticoagulant from heparin to nafamostat mesilate allowed dialysis to continue without rebleeding. These findings suggest that hemodialysis in patients with an AC associated with ADPKD may increase the risk of bleeding. Nafamostat mesilate may be useful in such cases.

Key words: autosomal dominant polycystic kidney disease, arachnoid cyst, subdural hematoma, hemodialysis, nafamostat mesilate

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Introduction

Autosomal dominant polycystic kidney disease (ADPKD) caused by a mutation in the PKD1 or PKD2 gene is a systemic disease consisting of renal and extra-renal manifestations. Arachnoid cysts (ACs) are an extra-renal manifestation associated with 8.1% of ADPKD patients (1) and can increase the risk of a subdural hematoma (SDH) (2). Although a SDH associated with an AC in patients with ADPKD has been infrequently reported (3, 4), the occurrence of intracystic hemorrhaging during hemodialysis (HD), which increases the risk of bleeding (5), remains unknown. To the best of our knowledge, this is the first report of dialysis-induced SDH in an AC associated with ADPKD.

Case Report

A 59-year-old woman was admitted to our hospital in November 2013 with a 2-month history of a physically disabled arm and leg on the right side and a 1-week history of a decreased level of consciousness. Her past medical history included ADPKD, hypertension and myocardial infarction. She had taken aspirin to prevent thrombogenicity. A family survey revealed renal failure caused by ADPKD in her father. She had a blood pressure of 143/93 mmHg and a pulse of 75 beats/min with a regular rhythm. A neurological examination showed a mild disturbance of consciousness (Glasgow coma scale: E4V4M6) and weakness of the face, leg and arm on the right side. Her laboratory data were as follows: blood urea nitrogen, 116.7 mg/dL; serum creatinine, 6.86 mg/dL; serum total protein, 7.5 g/dL; serum albumin, 3.4 g/dL; serum sodium, 137 mEq/L; serum potassium, 4.8 mEq/L; and blood glucose, 88 mg/dL. Plasma osmolality was 330 mOsm/kg H2O (normal value: 285-295). The patient’s platelet count (154 × 10^9/mL), prothrombin time (114 s), activated partial thromboplastin time (32.3 s) and fibrinogen level (4.73 g/L) were normal. Brain computed tomography...
Because it was strongly suspected that the SDH observed in the AC was associated with HD, nafamostat mesilate (40 mg/hr) was used as a substitute for heparin on the 17th and 20th days. Although no increase in the SDH was observed (Figure E), HD was briefly discontinued. On the 39th day, HD was reinitiated using nafamostat mesilate due to exacerbation of uremia. Because absorption of the hematoma was observed on brain CT (Figure F, G), heparin was used as the anticoagulant after the 56th day. The SDH disappeared on the 107th day without any rebleeding episodes (Figure H). The patient recovered well without neurological deficits.

Discussion

It has been suggested that ACs may predispose patients to subdural hygromas, intracystic hematomas and SDHs (6). Because the incidence of an AC in patients with ADPKD (8.1%) is ten times higher than in the general population (0.8%) (1), bleeding episodes associated with an AC are expected to be increased in patients with ADPKD. To the best of our knowledge, however, such cases have been reported in only four patients (3, 4), two of whom showed a SDH (CT) showed bilateral subdural hygromas with a left-side predominance (Figure A). Brain magnetic resonance imaging (MRI) revealed multiple ACs in the bilateral convexity, cisterna magna and quadrigeminal cistern. In addition, multiple liver and renal cysts were observed on chest and abdominal CT. The patient was diagnosed with end-stage renal disease (ESRD) due to ADPKD and received HD as follows: blood flow rate, 100 mL/min; heparin dosage, one-shot injection of 1,000 IU at the start of HD followed by a maintenance dose of 500 IU/hr thereafter; using a hemodialyzer, kf-m10® (Asahi Kasei, Tokyo, Japan). Although the patient’s impaired consciousness was rapidly corrected, she had a feeling of sickness suggesting dialysis disequilibrium syndrome. Her right hemiparesis disappeared with the normalization of plasma osmolality. On the 15th day after the initiation of HD, a follow-up brain CT for the bilateral subdural hygromas revealed a fresh SDH overlying the left frontal convexity of the cerebrum (Figure B). On brain MRI the following day, the SDH was identified as an intracystic hemorrhage (Figure C). A T2*-weighted image demonstrated an extremely clear low-signal change along the cyst wall (Figure D). Because it was strongly suspected that the SDH observed in the AC was associated with HD, nafamostat mesilate (40 mg/hr) was used as a substitute for heparin on the 17th and 20th days. Although no increase in the SDH was observed (Figure E), HD was briefly discontinued. On the 39th day, HD was reinitiated using nafamostat mesilate due to exacerbation of uremia. Because absorption of the hematoma was observed on brain CT (Figure F, G), heparin was used as the anticoagulant after the 56th day. The SDH disappeared on the 107th day without any rebleeding episodes (Figure H). The patient recovered well without neurological deficits.
adjacent to the AC (3). Although these ADPKD patients had hypertension without ESRD, major skull trauma or apparent blood coagulopathy were uncommon (3). In these patients, the hemorrhage might be facilitated by AC-relevant calvarial thinning and dural detachment. In addition, decreased cushioning due to a low-compliant cyst might facilitate bleeding from the bridging vein during minimal trauma (3).

In contrast to previous reports (3, 4), our patient showed impaired consciousness and transient right hemiparesis due to her hyperosmolar state related to ESRD associated with ADPKD (7). She had subdural hygromas at the initial visit, followed by an intracystic hemorrhage soon after the initiation of HD. Although the development of a SDH from a subdural hygroma is not uncommon (8), ESRD involving various coagulopathies (5) and the use of heparin during HD might facilitate the development of a SDH. In addition, fluctuations in the cerebral blood flow and intracranial pressure during HD (9) might facilitate tearing of the stretched bridging vein between the outer arachnoid membrane and dura, or in the cyst wall. Although the source of bleeding was not definitive, the low-signal change along the cyst wall on T2*-weighted imaging suggested a hemorrhage from the bridging vein located adjacent to the AC. Thus, an intracystic hemorrhage might be a critical complication at the initiation of HD in patients with an AC associated with ADPKD. In such cases, therefore, appropriate brain imaging should be performed and the patient should be provided with a thorough explanation in order to provide safe treatment.

In the present case, after the development of the SDH, the change of anticoagulant from heparin to nafamostat mesilate made it possible to continue HD without increased bleeding. This finding is consistent with a previous report that nafamostat mesilate might be beneficial to the recovery of damaged sites following the onset of a cerebral hemorrhage (10). The advantage of nafamostat mesilate might be due to the effect of a prolonged blood coagulation time limited to the extracorporeal circulation circuit (11), while with heparin there is a sustained systemic effect that lasts for several hours after withdrawal. Therefore, the use of nafamostat mesilate might be recommended in patients with an AC associated with ADPKD who are undergoing HD for the first time. In addition, the avoidance of potentially pathogenic circulatory volume shifts in HD and blood pressure control could reduce bleeding episodes (8). Although patients with ADPKD do better on HD, probably due to higher concentrations of erythropoietin and hemoglobin or lower comorbidity (2), careful precautions to prevent bleeding would further improve the outcomes of ADPKD patients.

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

References