Case Report

Metastatic intracranial subdural empyema from renal cyst infection in autosomal dominant polycystic kidney disease

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Introduction

Approximately 30–50% of patients with autosomal dominant polycystic kidney disease (ADPKD) develop renal infection [1]. Cyst infection and acute pyelonephritis are the most common infectious complications, although bacteraemia, perinephric and extrarenal abscess can also be seen. The problem is that eradication of cyst infections with conventional antibiotics is sometimes unsuccessful chiefly because of poor penetration of antibiotics into infected cysts. As a result, a significant portion of patients with ADPKD suffer from latent cyst infection, which may result in bacteraemia and/or metastatic extrarenal infection.

We describe here a case of ADPKD whose intracranial subdural empyema indicated a metastatic abscess from an uncontrolled cyst infection accompanying ADPKD.

Case

A 76-year-old woman was admitted for sudden onset of fever and altered mental status in June 2005. The patient had developed progressive renal dysfunction due to ADPKD since 1991. Although her renal function deteriorated to end-stage renal disease, she refused to start dialysis. She had repetitive episodes of renal cyst infection, for which she was hospitalized and treated with ciprofloxacin, only resulting in suboptimal improvement.

On admission, she was disoriented, with body temperature of 39.2°C, blood pressure of 164/100 mmHg, and heart rate of 160 due to paroxysmal supraventricular tachycardia. Physical examination revealed no meningeal signs. Her abdominal exam was normal except for mild right cost-vertebral angle (CVA) tenderness. Abnormal laboratory findings included white blood cell (WBC) count of 15,800/μl, haemoglobin of 9.6 g/dl, urea nitrogen of 57.2 mg/dl, creatinine of 5.14 mg/dl and bicarbonate of 17.2 mEq/l. Urinary analysis showed protein 3+ by dipstick; red blood cell of 51–100/high power field (hpf), WBC of 11–20/hpf, and granular casts 1+ by microscopy. The renal indices did not differ from those of previous tests and thus uraemic encephalopathy seemed unlikely. Blood culture was negative, but trace amounts of Escherichia coli were isolated in urine culture. Lumbar puncture revealed clear cerebrospinal fluid with a cell count of 12/mm³ and a negative culture test, indicating the absence of a demonstrable association with severe bacterial meningitis. Abdominal computed tomography (CT) revealed inflamed fatty tissues peripheral to the lower pole of the right kidney, suggesting the presence of an active cyst infection (Figure 1A). Head CT showed a left-sided crescent-shaped intracranial subdural fluid collection (Figure 2A), while no other abnormalities in the skull or sinuses were pointed out. She quickly lost the ability to articulate, open her eyes spontaneously or obey commands. Subsequently, CT scan revealed an enlargement of the intracranial fluid collection with mass effect. She underwent urgent surgery, and a brownish yellow fluid was drained from the subdural space. The fluid neither included haematoma nor bloody components. Colonies of Gram-negative rods phagocytosed by large amounts of leucocytes were identified by
microscopy, and *E. coli* were isolated from the drained pus culture.

After the patient was treated with ceftriaxone and phenytoin, her mental status gradually improved, to an extent where she was able to articulate and obey commands. On a head CT series, the subdural fluid collection decreased in size (Figure 2B), and inflammatory change of perirenal fatty tissue diminished (Figure 1B) as did bacteuria and pyuria. Her renal function, however, deteriorated progressively, and she started haemodialysis on a regular basis.

To determine whether *E. coli* isolated from the drained pus was identical to that isolated from the urine on admission, we performed two different analyses of these strains. First, *in vitro* microbiology sensitivity test demonstrated that both strains showed precisely the same minimum inhibitory concentration (MIC) against 15 different antibiotics. Second, after digestion of chromosomal DNA of both strains with restriction endonuclease, Xba1, restriction products were analysed by pulse-field gel electrophoresis (PFGE). PFGE profiles were analysed using GelCompar II software (Applied Maths BVBA, Belgium) and interpreted according to the guideline published previously [2]. To compare restriction patterns, we calculated percentage similarities with the Jaccard coefficient by the unweighted pair-group method using arithmetic averages. As a result, PFGE revealed indistinguishable Xba1 restriction patterns of both strains (Figure 3). These data demonstrated that the *E. coli* strain isolated from the drained pus was most probably identical from that isolated from the urine on admission.
Intracranial subdural empyema represents a closed infection between the outermost layer of the meninges, the dura, and the arachnoid. This intracranial infection is most frequently a complication of sinusitis, otitis or neurosurgical procedures [3]. However, in this case, these origins of subdural empyema were denied by history, physical examination and imaging studies. In addition, isolation of \( E.\) coli from drained empyema has been very rarely reported [4,5], which strongly suggested that the empyema was a metastasis from a sub-diaphragmatic site. Meanwhile, renal cyst infection is quite common in ADPKD but includes diagnostic problems [6]. In the present case, the patient had repetitive episodes of renal cyst infection and presented on admission with right CVA tenderness with pyuria and inflamed fatty tissue around the right cystic kidney on abdominal CT which indicated latent renal cyst infection. The fact that two strains of \( E.\) coli isolated from urine and empyema showed precisely the same antibiotic sensitivity strongly supported that the empyema was a metastatic renal cyst infection.

Furthermore, we performed PFGE; a well-established molecular sub-typing method in discriminating among bacterial strains. This method has been generally used for surveillance of bacterial outbreak, but recently it has been applied to analyse whether a particular strain was responsible for separate infections in the same individual [7]. The results clarified that in our case one particular \( E.\) coli strain was responsible for both the urinary tract infection and the extrarenal infections.

Bacteraemia or extrarenal abscess metastasized from urinary tract infections are potential complications of ADPKD. To our knowledge, however, there have been very few case reports of renal cyst infection associated with extrarenal infection [8] and this is the first case report of an empyema associated with renal cyst infection.

Advanced age and the female sex, as in this case, have been identified as possible predisposing factors of renal infection in ADPKD [9]. Renal cyst infection is notorious for its resistance to antibiotics. In our case, ciprofloxacin was administered repeatedly for renal cyst infection prior to this admission, but apparently this failed to eradicate the pathogen.

In summary, we report a case of ADPKD complicated with intracranial subdural empyema caused by \( E.\) coli. The link between these remote infections was confirmed by clinical pharmacological and molecular sub-typing methods of the responsible microorganism and suggest that the empyema was a metastatic renal infection. This case calls for the need for adequate control of urinary tract infections in ADPKD patients to avoid fatal metastatic infection.

**Conflict of interest statement.** None declared.

**References**


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