Fitz-Hugh-Curtis Syndrome Consequent to a Wound Infection Following Removal of a Peritoneal Dialysis Catheter

Fitz-Hugh-Curtis syndrome (FHCS) is a condition characterized by inflammation of the liver capsule (perihepatitis) and adjacent peritoneal surfaces. We report a case of FHCS developing in a peritoneal dialysis (PD) patient in whom catheter removal due to recurrent peritonitis was complicated by post-operative wound infection. To the best of our knowledge, this is the first case description of FHCS in the context of PD.

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KEY WORDS: Fitz-Hugh-Curtis syndrome; perihepatitis; peritoneal dialysis.

Fitz-Hugh-Curtis syndrome (FHCS) is a condition characterized by inflammation of the liver capsule (perihepatitis) and adjacent peritoneal surfaces. It has, typically, been described

in young women of child-bearing age in association with either overt or latent pelvic inflammatory disease (1). Previously thought to be primarily attributable to *Neisseria gonorrhoeae*, of late, cases of FHCS due to *Chlamydia trachomatis* infection have been shown to outnumber those due to *Neisseria* by a ratio of 5:1 (2). Although these organisms are regarded as the 2 major pathogens for the syndrome, other pathogens such as tuberculous and non-tuberculous mycobacteria have been reported (3,4). Bacterial transmission to the liver may occur through hematogenous or lymphatic spread and in women from an infected fallopian tube via the right paracolic gutter. We report a case of FHCS developing in a peritoneal dialysis (PD) patient in whom catheter removal due to recurrent peritonitis was complicated by post-operative wound infection. To the best of our knowledge, this is the first case description of FHCS in the context of PD.

CASE REPORT

A 58-year-old woman with end-stage renal disease due to diabetic nephropathy was commenced on continuous ambulatory PD. After 4 months, a mild exit-site discharge was noted, to which local gentamycin cream was applied. Cultures grew methicillin-sensitive *Staphylococcus aureus* and the patient was then administered oral cephalexin. Despite this treatment, the exit-site infection proved to be refractory and actually progressed to clinically evident tunnel infection (manifested by tenderness over the tunnel tract). Catheter removal was advised but the patient refused. A 4-week course of intraperitoneal (IP) cefazolin was given with good response, that is, resolution of the exit-site/tunnel infection.

A month later, she presented with diffuse abdominal tenderness and a cloudy effluent with a white blood cell (WBC) count of 7,150/ μ L of which 86% were polymorphs. Coagulase-negative *Staphylococcus* was cultured and successfully treated with IP cefazolin. Eight weeks later, coagulase-negative *Staphylococcus* peritonitis recurred and this time responded well to IP vancomycin. An ultrasound examination of the catheter tunnel revealed a small fluid collection around the internal cuff. Under vancomycin coverage, the catheter was removed and the patient transferred to hemodialysis. Post-operatively, a wound infection with abscess formation developed. The abscess was drained and cultures grew AmpC-resistant *Enterobacter aerogenes*. Treatment with intravenous ciprofloxacin resulted in eradication of the wound infection.

Four weeks later, she complained of severe pleuritic type pain in the right upper abdominal quadrant. On examination, she was afebrile with marked tenderness over the area. The former exit-site and tunnel wound were healed. Laboratory data showed a WBC count of 8,500/ μ L and an increased C-reactive protein (CRP) level of 11.7 mg/dL (normal range 0.0 – 0.5). Transaminases and lactic dehydrogenase (LDH) were within normal limits. A chest X ray was of normal appearance. Abdominal ultrasound was non-contributory apart for the fact that it showed no signs of cholecystitis. Computed tomography (CT) of the abdomen in the arterial

phase revealed linear hepatic capsular enhancement and in the portal phase perihepatic fat stranding (Figure 1, 2). Based on the clinical picture and CT findings, a diagnosis of FHCS was made. The patient was then referred for a gynecological examination, in which pelvic inflammatory disease was excluded. She was treated with a 3-week course of ciprofloxacin with complete resolution of her symptoms and CT findings.

DISCUSSION

Fitz-Hugh-Curtis Syndrome refers to inflammation/infection of the liver capsule without involvement of the liver parenchyma. It has mainly been described in pre-menopausal women in association with pelvic inflammatory disease. The proposed mechanism in this setting is presumed to be

Figure 1 — Arterial phase CT scan shows linear enhancement at lateral surface of right hepatic lobe. CT = computed tomography.

Figure 2 — Portal phase CT scan shows perihepatic fat stranding (arrow). CT = computed tomography.

ascending transperitoneal bacterial spread from the pelvis along the right paracolic gutter although hematogenous and lymphatic pathways may also be implicated. Anecdotally, male patients have also been reported (5). As its synonym, gonococcal perihepatitis, suggests, *Neisseria* was thought to be the primary offending pathogen in the 1970s. Recent literature has, however, pointed to *Chlamydia* as the most common pathogen (2).

The principal clinical manifestation of FHCS is intense pleuritic pain in the right upper quadrant which may be referred to the right shoulder. Of the laboratory parameters, a retrospective study reporting the clinical features of FHCS in 25 patients found an elevated erythrocyte sedimentation rate and CRP in 76 and 92% of cases, respectively (6). White blood cell count was increased in 60%. In keeping with this data, our patient's CRP was markedly elevated. This non-specific clinical scenario of FHCS mimics, in particular, either biliary tract or pulmonary disease and may thus lead to the diagnosis being missed or delayed. This is particularly true when signs of pelvic inflammatory disease are absent. The diagnosis, as in our case, was established by the finding of hepatic capsular enhancement on the arterial phase of a contrast-enhanced CT. This enhancement of the liver capsule is attributed to an increased blood flow or inflammation of the capsule. It has been reported as a characteristic feature of the acute phase of FHCS (7). However, it is a non-specific finding, as it may be seen in other conditions such as acute cholangitis, peritoneal carcinomatosis, and systemic lupus erythematosus (8). Violin string-like adhesions between the liver and the parietal peritoneum have also been described as a typical finding of FHCS, especially in the chronic phase of the disease. They may be rarely visualized on sonography or CT (7) and are considered a definitive diagnostic feature of FHCS on laparoscopy.

To our knowledge, our case is the first report of FHCS in the context of a patient who was treated with PD. It is, as well, the first time that AmpC *Enterobacter* has been documented as the likely causative organism. Treatment of the syndrome consists of appropriate antibiotic coverage. As such, our patient responded to ciprofloxacin to which the AmpC *Enterobacter* was sensitive. In cases of refractory pain, laparoscopic lysis of adhesions may be performed.

DISCLOSURES

The authors have no financial conflicts of interest to declare.

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