Case report: The first case of Achromobacter xylosoxidans-related tunnel infection in a patient receiving peritoneal dialysis

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Abstract
Rationale: Achromobacter xylosoxidans infection is mostly reported in immunocompromised patients. Until now, it is still rarely reported in patients undergoing peritoneal dialysis.

Patient concerns: This is the 1st case of A xylosoxidans infection due to tunnel infection of a Tenckhoff catheter.

Diagnosis: The diagnosis was confirmed by the report of culture.

Interventions: Risk factors for this infection in peritoneal dialysis include uremia with an immunocompromised state, contamination due to inexperienced skills, and aqueous environment of the dialysate.

Outcome: We believe that finding the source of A xylosoxidans contamination is the most important aspect of the overall treatment of the infection.

Lessons: Environmental investigation of suspected source contamination is warranted in those with A xylosoxidans infection. Once the diagnosis is made, removal of the Tenckhoff catheter should not be delayed.

Abbreviations: AX = Achromobacter xylosoxidans, PD = peritoneal dialysis, WBC = white blood cell.

Keywords: Achromobacter xylosoxidans, peritoneal dialysis, tunnel infection

1. Introduction

Achromobacter xylosoxidans (AX) infection is very rare in patients undergoing peritoneal dialysis (PD). It is mostly detected in immunocompromised patients, such as patients undergoing chemotherapy. Until now, there were only 12 previously reported cases[1–10] of patients receiving PD contracting AX infection. Two were an exit site infection[2] and the other 10 cases were peritonitis. Herein, we report the 1st case with AX infection due to tunnel infection of a Tenckhoff catheter. We also provide a review of literatures on AX infection in PD patients.

2. Case report

A 45-year-old woman had been receiving PD since 5 years ago due to immunoglobulin A nephropathy, and she had no other disease. Her dosage of dialysate was 1.5% of 2L Dianel solution for 4 bags per day. She had never experienced any PD-associated infection in the past 4 years. One year before her 1st hospitalization, she noticed 2mL of pus around the exit site. After a repeated course of training in PD procedures, symptoms and signs of exit site infection improved. She was afebrile at the time and her dialysate was not cloudy. The pus culture report yielded Pseudomonas aeruginosa, which was sensitive to many antibiotics (Gentamicin, Amikacin, Carbapenem, Ceftriaxone, Cefpime, Ciprofloxacin, Piperacillin/Tazobactam, and Colistin). Therefore, she received oral Ciprofloxacin for 2 weeks, and the exit site became dry and clean. One month before her 1st hospitalization, she complained of subcutaneous pain along with the Tenckhoff catheter over her left abdomen. There was again no cloudy dialysate and no pain over other abdominal region. She was then hospitalized and received intravenous Amoxycillin/Clavulanate for 2 weeks. However, her condition did not improve. Thus, the surgeon removed the left Tenckhoff catheter and closed the fascia with 2 layers (posterior and anterior sheath). A new Tenckhoff catheter was inserted via her right abdomen. Her dialysate was clean without any turbid materials at the perioperative period. Later, the culture of the removed catheter grew AX. This AX was sensitive to the following antibiotics, Ampicillin/Sulbactam, Trimethoprim/Sulfamethoxazole, Carbapenem, Cefoperoxazone 500mg/Sulbactam 500mg, Ceftazidime, Piperacillin/Tazobactam, and Tigecycline. This culture was identified by an automated identification system, VITEK 2 system (bioMérieux-Vitek, Hazelwood, MO). She was discharged with oral 3rd-generation Cephalosporin daily and continued to use the new Tenckhoff catheter for PD after discharge. Two weeks after the operation, she was admitted again due to acute onset of abdominal pain along with cloudy dialysate for 3 days. Her white blood cell (WBC) counts of dialysate were 3190/μL. Other examinations of dialysate were as follows: 120...
U/L of lactate dehydrogenase, 0.35 g/dL of total protein, and 450 mg/dL of glucose. Her blood tests were 12,000/cumm of WBC, 89 mg/dL of blood urea nitrogen, 12.2 mg/dL of serum creatinine, 3.5 mg/L of C-reactive protein, 110 mg/dL of blood sugar, 120 mg/dL of low-density lipoprotein cholesterol, and 200 mg/dL of triglyceride. Her blood and urine cultures were both negative. Therefore, PD-related peritonitis was diagnosed.

Empirical antibiotics with 1st- and 3rd-generation Cephalosporins (Cefazolin and Ceftazidime, 125 mg/L) were infused via irrigation. Three days after the infusion of antibiotics, the WBC of dialysate dropped to 1729 and 169/μL, and the dialysate became clean and colorless. Finally, the culture of dialysate yielded AX. The antimicrobial susceptibility testing of AX during this 2nd admission was the same as 2 weeks ago from the removed catheter suggesting the same pathogen as the cause. The test showed that the pathogen was resistant to 1st-generation Cephalosporin and Ciprofloxacin, but sensitive to Ceftazidime and Ampicillin/Sulbactam. The abdominal tomography disclosed dirty fat plane over the previous tunnel (Fig. 1B), compared with the previous a year ago (Fig. 1A). Finally, the final diagnosis was inadequate treatment for AX-related tunnel infection, which progressed to PD-related peritonitis. Since the dialysate became clean and there was no more abdominal pain, she was discharged with oral Ampicillin/Sulbactam for 2 more weeks. The total 3-week antibiotics courses were applied and she recovered well. This study had been approved by the patient herself and she signed the informed consent.

### 3. Discussion

*Achromobacter xylosoxidans* (formerly *Alcaligenes xylosoxidans*) is a nonfermenting, aerobic, oxidase- and catalase-positive, and gram-negative rod with peritrichous flagella. Normally, it is distributed in aqueous environments including soil, water, rotten eggs, and dairy products. In human bodies, it may also exist as normal flora over skins and in the gastrointestinal tract. However, in immunocompromised patients such as patients undergoing chemotherapy, it could cause pneumonia, pharyngitis, catheter-related infection, external otitis, and urinary tract infections. Until now, there were only 12 reported cases of AX-related infection in PD [1–11] (Table 1). Of them, most (10 cases) were PD-related peritonitis, and 2 were an exit site infection.[2,11] Our case is the 13th case of AX-related infection in PD but it is the 1st case with AX-related PD tunnel infection.

| Case, year | Age, gender | Causes of uremia | Possible clues to infection | Outcome |
|------------|-------------|------------------|----------------------------|---------|
| Peritonitis (n=11) |
| 1 [1], 1980 | 53, M | Diabetic nephropathy | 5th peritonitis | Cured |
| 2 [2], 1984 | 40, M | Acute tubular necrosis | 1st peritonitis, but many time bacteremia | Expired |
| 3 [3], 1986 | 34, F | Glomerulopathy | 1st peritonitis | Cured |
| 4 [4], 1995 | 45, M | Glomerulopathy | Unknown | Catheter removed |
| 5 [5], 1998 | 52, F | Diabetic nephropathy | 2nd peritonitis; blindness | Catheter removed |
| 6 [6], 2001 | 46, F | IgA nephropathy | 2nd peritonitis | Catheter removed |
| 7 [7], 2001 | 35, F | IgA nephropathy | 1st peritonitis; graft failure | Catheter removed |
| 8 [8], 2004 | 16, M | Idiopathic Fanconi syndrome | 7th peritonitis, nonaseptic technique | Catheter removed |
| 9 [9], 2010 | 51, F | Diabetic nephropathy | 5th peritonitis, cirrhosis | Catheter removed |
| 10 [10], 2012 | 31, M | Obstructive uropathy | 1st peritonitis | Cured |
| Exit site infection (n=2) |
| 11 [11], 2012 | 82, F | Diabetic nephropathy | 1st exit site infection | Cured |
| 12 [12], 2013 | 70, F | Unknown | polymicrobial exit site infection | Cured |
| Tunnel infection (n=1) |
| 13, 2016 (this case) | 45, F | IgA nephropathy | 1st tunnel infection, 1st peritonitis | Catheter removed |
| Subtotal | 46.2, F (61.5%) | Diabetic nephropathy: 33.3%; glomerulopathy: 41.6% | 1st infection: 50% | Expire: 7.7%; catheter removed: 53.8% |

Figure 1. (A) Abdominal tomography for tunnel infection. There is no infection over the new Tenckhoff catheter (white arrow) over her right abdomen, but tunnel infection can be detected over the previous removed Tenckhoff catheter with dirty fat plane (white arrow head). (B) Abdominal tomography 1 year ago. Clean fat plane near the Tenckhoff catheter over left abdomen (white arrow).
the PD cases were due to glomerular disease, followed by diabetic nephropathy (33.3%). Most cases (53.8%) recovered after the removal of catheters. This was because AX may form biofilm on plastic materials like PD catheters and was difficult to be eradicated without the removal of catheters. Keeping PD catheters and manually flushing them could mechanically shear the biofilm and cause detachment of cells or aggregates. Therefore, we suggested strict removal of PD catheters immediately if a diagnosis of AX-related infection was made.

The virulence of AX is considered to be weak, so it commonly infects immunocompromised patients. There are some characteristics for PD favoring the growth of AX. Firstly, renal failure confers the immunocompromised state. Secondly, glucose containing dialysate provides an aqueous environment with much nutrition for AX growth. As in case 9, cirrhosis also made a more immunocompromised state. However, we believed that there are still some reasons predisposing AX infection in PD because reports of AX infection in PD were still rare. One reason was the inexperienced skills for PD (such as case 5 with blindness). Half of the AX infections in PD were more than 1 peritonitis (5 times peritonitis for 2 cases and 7 times peritonitis for 1 case). Finally, AX contaminated water without thorough disinfection was also the major problem. We should bear in mind the possibility of contaminated sources if the occurrence of AX-related peritonitis was in more than 1 patient. Unfortunately, there was no clear source of AX infection in 13 cases, except for our patient. We performed environmental investigation cultures from available open solutions in her house. Then a positive culture for AX was detected in a tap water sample from her faucet. According to the antibiotic susceptibility, AX from her PD catheter and her dialysate may came from the tap water from the faucet. Our patient claimed that she did not regularly wash her hands thoroughly with disinfectants. She also admitted to touching her PD catheter after washing hands with tap water without using disinfectants. That can explain why she had AX-related tunnel infection, followed by AX-related peritonitis. This is the 1st case of AX-related PD infection with clear environmental investigations. Our investigation helps to understand the mechanisms of AX-related PD infection. It provides a better understanding of AX in the environments causing infection in PD.

4. Conclusion
AX-related infection is still rare currently but we still should be reminded to remove PD catheters if this diagnosis is confirmed. Meticulous environmental investigation can guide clinicians to avoid recurrent AX infection. Despite its rarity, we should not delay the diagnosis of this disease, especially in high-risk patients.

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