A Rare Case of Pyonephrosis in an Infant Induced by Extended-Spectrum Beta-Lactamase-Producing *Klebsiella pneumoniae*

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**Abstract**

Pyonephrosis is a rare condition in both adult and pediatric populations. Here, the author presents a rare case of pyonephrosis induced by extended-spectrum beta-lactamase-producing *Klebsiella pneumoniae* in a 12-month-old girl presenting with a picture of urosepsis. The patient presented with febrile urinary tract infection and was unresponsive to intravenous meropenem. Physical examination revealed huge, firm and irregular right renal swelling. Ultrasound and computed tomography imaging revealed severely hydronephrotic right kidney, and laboratory investigations showed elevated C-reactive protein level (22.9 mg/dl). Emergency percutaneous nephrostomy tube was inserted, pus was drained (20 ml) and intravenous vancomycin and amikacin were started. Her general condition improved, and urine culture was negative. Functional assessment with dimercaptosuccinic acid renal scan revealed that the split renal function was 5% on the right and 95% on the left side, and the bladder outline was smooth with no reflux in voiding cystourethrogram. A right nephrectomy was done a week later using the anterior subcostal approach. The postoperative course was smooth. Histopathological examination was diagnostic for xanthogranulomatous pyelonephritis. No adverse events were reported in the follow-up over 12 months. It can be concluded that a high degree of suspicion, rapid initiation of appropriate antibiotics and drainage of pus are crucial in the management of pyonephrosis.

**Keywords:** Extended-spectrum beta-lactamase, infant, infection, pyonephrosis, sepsis

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**Submitted:** 29-Jun-2018  **Revised:** 03-Jan-2019  **Accepted:** 07-Feb-2019  **Published:** 17-Apr-2020

**INTRODUCTION**

Pyonephrosis is an infection that leads to obstructed hydronephrotic kidney, with supplicative destruction of parenchyma and pus accumulation, and usually progresses to total or near-total loss of renal function. Pyonephrosis is rare in both pediatric and adult populations. Treatment with potent intravenous antibiotics are highly effective, provided urgent decompression and drainage of pus are achieved. Here, a rare case of pyonephrosis in an infant caused by extended-spectrum beta-lactamase (ESBL)-producing *Klebsiella pneumoniae* is presented. To the best of the author’s knowledge, this is the first such case being reported from the Kingdom of Saudi Arabia.
CASE REPORT

A 12-month-old girl with an unremarkable antenatal, natal and postnatal history presented with febrile urinary tract infection (UTI) caused by ESBL-producing *K. pneumoniae*. Intravenous meropenem (20 mg/kg every 8 h) was started, but the patient was unresponsive and showed signs of clinical deterioration. Ultrasound and computed tomography scan showed severely hydronephrotic right kidney filled with purulent fluid. It extended from the right upper quadrant to right iliac fossa and crossed the midline [Figures 1 and 2]. The patient appeared ill, pale, dehydrated and lethargic, had a temperature of 40°C, blood pressure of 100/60 mmHg and was tachycardic (150 beats/min). Physical examination revealed huge, firm and irregular right renal swelling. Laboratory investigations showed leukocytosis (white blood cell count: 22 K/μl), anemia (hemoglobin level: 7 g/dl) and thrombocytosis (platelet count: 998 K/μl). C-reactive protein was elevated (22.9 mg/dl). Other laboratory investigations including liver and renal function tests as well as prothrombin time and partial thromboplastin time tests were normal.

Emergency percutaneous nephrostomy tube was inserted, and 20 ml of pus was drained. Intravenous vancomycin (15 mg/kg every 6 h) and amikacin (2.5 mg/kg every 8 h) were started. The patient’s general condition improved, and the temperature returned to normal the next day. Urine culture obtained from the nephrostomy tube was negative. Despite remarkable improvement, platelet count continued to increase for 1 week after the nephrostomy tube insertion, reaching 1490 K/μl. The patient was also started on aspirin 5 mg/kg every other day together with ferrous sulfate to help optimize hemoglobin level. She was maintained on oral trimethoprim–sulfamethoxazole prophylaxis (2 mg/kg once daily) to prevent recurrence of infection until definitive management was carried out. Dimercaptosuccinic acid renal scan was done at 6 weeks of treatment initiation and showed a split renal function of 5% on the right side and 95% on the left [Figure 3]. The bladder outline appeared smooth with no reflux in voiding cystourethrogram. There was improvement in the serum hemoglobin (11 g/dl) and a drop in the serum platelet levels (500 K/μl).

Based on the clinical presentation and the results of the renal scan, right nephrectomy was done a week later using the anterior subcostal approach. There were extensive adhesions between the kidneys, Gerota’s fascia and peritoneum. The right ureter was identified and followed to the ureteropelvic junction. Identification of the renal hilum was difficult. Careful dissection was carried out until the renal artery/vein was identified and ligated. The patient had a smooth postoperative course and was discharged 2 days after the surgery with no prescribed medication. Histopathological examination using hematoxylin and
eosin stain revealed marked reduction in renal tubules and glomeruli, increased interstitial tissue, thick-walled blood vessels, heavy interstitial mononuclear inflammatory cell infiltration and sheets of foamy histiocytes [Figure 4]. Immunohistochemical staining for CD68 was positive for histiocytes [Figure 5]. Subsequently, a diagnosis of xanthogranulomatous pyelonephritis was made. The patient was followed up at 3, 6 and 12 months, with no reported adverse events.

**DISCUSSION**

Infectious and inflammatory reactions associated with pyonephrosis result in a spectrum of abnormalities. These range from simple infected hydronephrosis with virtually unimpaired renal function to xanthogranulomatous pyelonephritis, where the renal function is severely impaired.[3] This can result in an increase in the bacterial load and pus accumulation in the collecting system, as was the case in our patient. It is easier to diagnose older children with pyonephrosis, as they usually present with high-grade fever, chills and flank pain. On the other hand, diagnosis of pyonephrosis in neonates and infants is more difficult because of rarity of cases in this age group, limited knowledge and the nonspecific clinical presentation of palpable abdominal mass, UTIs and urosepsis, as was the case in our patient.[3]

Routine antenatal ultrasound screening is a valuable tool for early detection of various uropathies, especially when done by an experienced radiologist/obstetrician, to ensure congenital anomalies are not missed because they can lead to serious events postnatally. Prophylactic antibiotic administration initiated in the immediate postnatal period reduces the risk of a secondary UTI, at least until radiological workup and diagnosis are reached. Irrespective of the prophylactic treatment, infants with antenatally detected hydronephrosis are 12 times more likely to be hospitalized for pyelonephritis in the 1st year of life than those without it.[4]

Nephrectomy used to be performed for all cases of pyonephrosis on an emergency basis, but it had high morbidity and mortality.[1] Currently, as we did for our patient, nephrostomy is initially performed to drain the pyonephrotic kidney because it controls infection and reduces the risk of mortality from septicemia as well as is important for revealing the anatomy of the obstructed kidney.[6,7] Nephrostomy is then followed by imaging for the assessment of the degree of structural damage and split function, and a reconstructive surgery or nephrectomy is carried out, depending on the degree of renal damage.[8] It should be noted that nephrostomy can lead to complications, such as bleeding, sepsis, catheter blockage and dislodgment.

Appropriate antibiotics are crucial in pyonephrosis treatment,[8] given that analysis of cultures obtained from nephrostomies, bladder and blood in a study done by Ng et al.[6] showed that 70% of patients had positive cultures from at least one specimen and that 8% had more than one organism. The most common microorganisms isolated were *Escherichia coli* (30%), *Klebsiella* (19%), *Proteus* (8%), *Pseudomonas* (5%), *Enterococcus* (5%) and *Candida* spp. (5%).[2] Our patient had ESBL-producing *K. pneumoniae*. ESBL, an enzyme produced by Gram-negative bacilli, plays a role in increasing antibiotic resistance.[9] In a meta-analysis published by Flokas et al.,[10] the prevalence of ESBL was 5% among community-acquired and 12% among hospital-acquired infections. ESBL-producing strains are resistant to all penicillin as well as the majority of cephalosporins and aztreonam.
Regional knowledge regarding the most common microorganisms and their resistance pattern helps determine the most appropriate management protocol. In a study from the Aseer region of Saudi Arabia, Alamri et al.\textsuperscript{[11]} found that Gram-negative bacilli (\textit{E. coli} and \textit{K. pneumoniae}) are the most common uropathogens causing UTI and that the majority are resistant to antibiotics commonly used in clinical practice, as in our case. Therefore, in such cases, the author recommends using vancomycin, daptomycin and linezolid for the empirical treatment of UTI.

CONCLUSION

Pyonephrosis is a serious condition that can result in significant morbidity and mortality if not appropriately managed. Infection with a multidrug-resistant pathogen can further complicate and delay management. Clinical course of pyonephrosis reported in this case was caused by an ESBL-positive organism. Being vigilant with rapid initiation of appropriate antibiotics and drainage of pus was a crucial step in management.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient’s parents have given their consent for her images and other clinical information to be reported in the Journal. The patient’s parents understand that her name and initial would not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Peer review

This article was peer reviewed by three independent and anonymous reviewers.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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