CASE REPORT

Primary Renal Aspergillosis and Xanthogranulomatous Pyelonephritis in an Immuno-Competent Toddler

Durre Shohab[1], Ijaz Hussain[1], Athar Khawaja[1], Imran Jamil[1], Nazar Ullah Raja[2], Faizan Ahmed[1] and Saeed Akhter[1]

ABSTRACT

Aspergillosis is primarily a pulmonary disease so that renal aspergillosis is usually secondary to hematogenous spread from lungs. Primary renal aspergillosis, though a rare entity, is still seen in immuno-compromised individuals. Renal aspergillosis may lead to formation of focal abscesses, fungal bezoars and may cause ureteric obstruction. Treatment involves stabilization of patient and removal of fungal bezoars along with administration of anti-fungal agents. This report describes the case of localized primary renal aspergillosis with fungal bezoar formation in 2 years old immuno-competent child who presented in sepsis and acute renal failure and was successfully managed by nephroscopic removal of fungal bezoar and intravenous voriconazole. The other kidney required nephrectomy for xanthogranulomatous pyelonephritis.

Key Words: *Fungal bezoar. Primary renal aspergillosis. Voriconazole. Acute renal failure. Nephroscopy. Xanthogranulomatous. Pyelonephritis.*

INTRODUCTION

Aspergillosis is primarily a pulmonary pathology with hematogenous dissemination to other organs in persons with compromised immune status, such as diabetics, those on corticosteroids and HIV positive individuals or chronic alcoholic patients who had prolonged catheterization.[1,2] Primary renal aspergillosis is extremely rare in immuno-competent individuals.

The author hereby describe case report of a 2 years old immuo-competent child who developed primary renal aspergillosis with fungal bezoar, which was successfully removed by percutaneous nephroscopy.

CASE REPORT

A 2 years old male child presented with high grade fever, gross abdominal distention and anuria. His illness dated back to past one month when he had developed anuria and high-grade fever and was hospitalized in a local hospital. His investigation at that time showed serum creatinine level of 2.3 mg/dl and TLC (total leukocyte count) of 18000/µL. Ultrasound sonography (USG) showed gross right hydronephrosis and shrunken small hydronephrotic left kidney. Bilateral PCN (percutaneous nephrostomy) were done. There was adequate urine output from the right PCN but the left PCN drained just 50 - 100 cc of pus. Patient was improved with normalization of clinical and laboratory parameters. Urine and blood bacterial cultures showed no growth. No fungal culture was sent.

After stabilization, bilateral nephrostogram was done, showing no distal obstruction. Both PCN were removed and left DJ stenting was done. The child remained asymptomatic for 3 - 4 days but again developed high-grade fever with complete anuria. He was again hospitalized and started on antibiotics. He had high TLC and normal RFTs (renal function test). CT urogram showed right sided hydronephrosis, dilated ureter and a dense lesion in the right renal pelvis. Left kidney was small, hydronephrotic and shrunken. Right DJ stenting was done. Patient improved again followed by recurrent episodes of similar symptoms each time relieved by the change of DJ stent. In two weeks time, the patient underwent three time change of right DJ stent.

He presented to the department on the second post-operative day of the third change of the right DJ stent. He had high-grade fever, complete anuria and gross abdominal distention. On examination, patient had a pulse rate of 150/minute, temperature of 103°F and respiratory rate of 26 breaths per minute. Abdomen was grossly distended, tense and tender.

Patient was admitted and resuscitated. Urine cultures for both fungus and bacteria were sent. Investigation showed TLC of 22500/µL and creatinine of 4.0 mg/dL. USG showed gross ascites, shrunken hydronephrotic left kidney, normal size right kidney with raised echogenicity along with an echogenic mass in the right renal pelvis with no posterior acoustic shadowing. X-ray chest was normal.

Right sided PCN was done and intraperitoneal drain was placed which drained intraperitoneal fluid. Samples were directly taken from renal pelvis and sent for culture. Post-operatively patient was shifted to ICU for one day.

Department of Urology and Kidney Transplant[1] / Microbiology[2], Shifa International Hospital, Islamabad.

Correspondence: Dr. Durre Shohab, Department of Urology and Kidney Transplant, Shifa International Hospital, Sector H-8/4, Islamabad.
E-mail: capitalian232@yahoo.com

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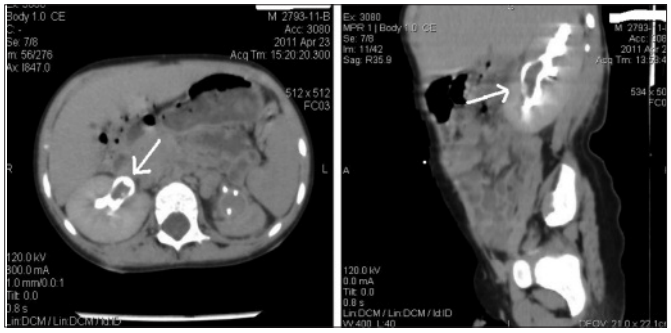

Figure 1: CT-Urogram delayed films showing the filling defect in the right renal pelvis (arrow).

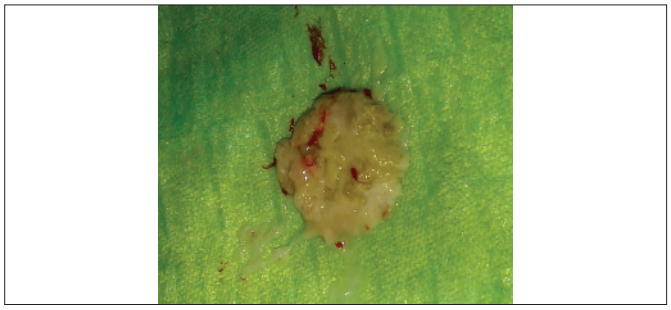

Figure 2: Fungal ball/bezoar removed by nephroscopy.

There was marked improvement in patient condition both clinically and biochemically.

Three days later right RUPG (retrograde uretero-pyelogram) was done; old DJ stents were removed. RUPG showed leakage from upper calyx. Patient continued to improve; there was adequate output from right PCN, peritoneal drainage gradually decreased and clinical and laboratory parameters normalized. At this stage culture report was received showing *Aspergillus fumigatus*. The renal pelvic mass was then suspected to be a fungal bezoar. Patient was started on voriconazole. Finally, percutaneous nephroscopic removal of fungus bezoar and right DJ stenting was carried out. Intraperitoneal drain was removed followed by right PCN and child was discharged in completely stable condition.

Three weeks later, DMSA (Dimercaptosuccinic acid) scan was done which revealed completely non-functioning left kidney. Suspecting left kidney as the probable source of infection left nephrectomy was done. Per-operatively, the kidney was full of pus. However, culture did not reveal any aspergillosis but histo-pathology showed xanthogranulomatous pyelonephritis. Currently, the child is totally asymptomatic with normal RFTs.

DISCUSSION

Ascending infection from the lower tract and *Aspergillus* cast in renal pelvis are the other two means of renal involvement by aspergillosis apart from hematogenous spread from lungs.[3] Primary renal aspergillosis, though a rare entity, is still seen in immuno-compromised individuals. A large case series revealed that *Aspergillus* infection of the kidney accounts for 30% (27/90) of total renal fungal infection and majority (63%) results from disseminated infection.[3] Primary urinary tract infections caused by aspergillosis affecting kidneys and other genitourinary organs are rare and are usually a part of systemic spread of aspergillosis.[3] Aspergillosis can also involve other genitourinary organs but this is rare and literature is limited to case reports such as those case of prostatic aspergillosis.[6] However, in a case series reported by Karim *et al*. out of 17 immunocompetent patients suffering from different forms of invasive aspergillosis no case of genitourinary aspergillosis was found.[7]

Aspergillosis of the kidney can lead to formation of multiple focal abscesses, *Aspergillus* cast of the renal pelvis, and ascending panurothelial aspergillosis of the urethra, bladder, pelvis and kidney.[2] There are also case reports of ureteric obstruction due to aspergillosis.[4,6]

Aspergillosis of urinary tract must be diagnosed promptly and treated aggressively. In untreated cases, it can cause urinary obstruction through formation of fungal balls resulting in hydronephrosis, oliguria or anuria, destruction of the renal parenchyma, ultimately sepsis and wide-spread dissemination of the organism, and death of the patient.[2]

Treatment includes relief of obstruction by removal of fungal ball along with antifungal therapy. Voriconazole is considered the gold standard for systemic antifungal treatment. It has been proved to be superior to amphotericin-B in terms of response and improved survival with fewer severe side effects.[5,8] Liposomal amphotericin-B is now considered a safe alternative to voriconazole.[8] Caspofungin, amphotericin-B lipid complex or posaconazole are considered the second line agents.

This case is unique in the sense that the patient was immuno-competent and just 2 years of age. Owing to extreme rarity of this disease in immuno-competent individual of this age group, the child was not diagnosed earlier and suffered prolonged morbidity. Even all the cultures sent earlier were bacterial not fungal. Also there was no evidence of systemic spread or satellite locus of disease. After the culture diagnosis, the authors were able to successfully manage the patient by percutaneous nephroscopic removal of bezoar followed by oral voriconazole.

Keeping in view the current case report, this disease can be expected to occur even in immuno-competent individuals even in this age group.

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