CASE REPORT

Fungal Pneumaturia: A Case Report on a Large Urinary Bladder Fungal Bezoar in a Young Diabetic Male

Charles Anthony C. Gaston, MD; Michael John Francis V. Gaston, MD, FPUA and Marie Carmela M. Lapitan, MD, FPUA

Division of Urology, Department of Surgery, Philippine General Hospital, University of the Philippines Manila

Fungal bezoars or fungus balls are extremely rare cases especially when they occur within the urinary tract. Reported here is a 26-year old diabetic male presenting with pneumaturia, passage of debris per urethra and lower urinary tract symptoms. He was initially managed as a case of enterovesical fistula. Further work-up revealed a urinary bladder fungal bezoar. The patient was managed by endoscopic morcellation and evacuation of the fungal ball from the bladder and anti-fungal therapy. Awareness of this rare clinical entity and its presentation will aid in its proper diagnosis and management.

Keywords: fungal pneumaturia, fungal bezoar, lower urinary tract symptoms

Introduction

Fungal bezoars or fungus balls are formations of fungal masses within pre-formed body cavities and usually occur in immunocompromised hosts. These are rare cases, particularly when they occur in the urinary tract, of which less than 20 cases have been reported.[1] Previous literature shows that fungal bezoars usually occur in elderly, immunocompromised patients, usually resulting in lengthy hospital stays and prolonged indwelling urinary catheters.[1,2] The few reported cases presented primarily with signs and symptoms associated with urosepsis and obstructive uropathy highlighted by fever, flank pain and azotemia.[2] Reported here is an unusual presentation of a large urinary bladder fungal bezoar in a 26-year old diabetic male, which initially presented with pneumaturia, passage of debris per urethra and lower urinary tract symptoms, without signs of sepsis.

The Case

A 26-year-old male, undiagnosed diabetic, presented with a 3-month history of pneumaturia and lower urinary tract symptoms. He eventually went into acute urinary retention and presented to the emergency department. A urethral catheter was inserted draining turbid urine with yellowish-white debris. Vital signs were normal, and the patient did not present with an acute abdomen. Aside from the palpably distended urinary bladder, the rest of the physical examination was unremarkable. Due to the history of pneumaturia and the character of debris drained per catheter, the patient was initially assessed to have an enterovesical fistula.

Laboratory examinations showed no leukocytosis on complete blood count (WBC $6.8x10^9$ cells/L). Serum creatinine was noted to be elevated at 148 umol/L. Urinalysis yielded a pH of 5.0 and showed numerous hyphal elements and yeast cells. There was also note of glucosuria (28 mmol/L) and microscopic pyuria (1093/HPF) as well on urinalysis. Presence of the glucosuria on urinalysis prompted random blood sugar checking which showed 294 mg/dL result. Capillary blood glucose monitoring was constantly elevated with a range of 220-350 mg/dL. Glycated hemoglobin turned out to be 10.1%. C-peptide was normal (2.5 ng/mL). The patient was diagnosed and managed as a case of early-onset, insulin- requiring, type 2 diabetes mellitus as well.

Given that diverticulitis is a major cause of enterovesical fistula, a contrast-enhanced computed tomography (CT) with intravesical contrast was ordered to search for it. The CT scan showed a urinary bladder filled with mottled soft tissue densities, admixed with air, for which fecaloid material was the primary consideration (Figure 1.A). However, no definite fistulous tract between the bowel and the urinary bladder was demonstrated. In addition, bilateral pelvocaliectasia with pockets of air in the right renal pelvis was seen in the CT scan. (Figure 1.B). Colonoscopy was performed but no fistulous tracts, masses or areas of erythema were noted. Eventually, the patient underwent cystoscopy which revealed a large amount of fibrinous materials occupying the entirety of the urinary bladder (Figure 2A).

Using the cystoscope, manual dislodging of the fibrinous material, morcellation, evacuation of the material using an Ellik evacuator was done. Approximately 300 cc of fibrinous material was removed from the urinary bladder. After reconstitution, the mass was found to have an aggregate measurement of 8.0cm x 7.0cm x 5.0cm (Figure 2B).

Intra-operative cystogram after evacuation of the material in the urinary bladder showed normal bladder outline, absence of vesico-ureteral reflux, no extravasation of dye and non-visualization of the bowels (Figure 2C).

The slide review of the collected material showed abundant fungal elements (Figure 3). The pathology report confirmed the presence of Candida spp.

Post-operatively, the patient was able to void freely after catheter removal. There was full resolution of his lower urinary tract symptoms. Serum creatinine went down to a normal value of 90 umol/L. The patient completed a 14-day regimen of Fluconazole. He was discharged well and was advised to continue insulin therapy.

On follow-up, 1 month post-operatively, it was noted that there was full resolution of the pelvocaliectasia on KUB ultrasound. During this time, the patient was satisfied with the state of his voiding.

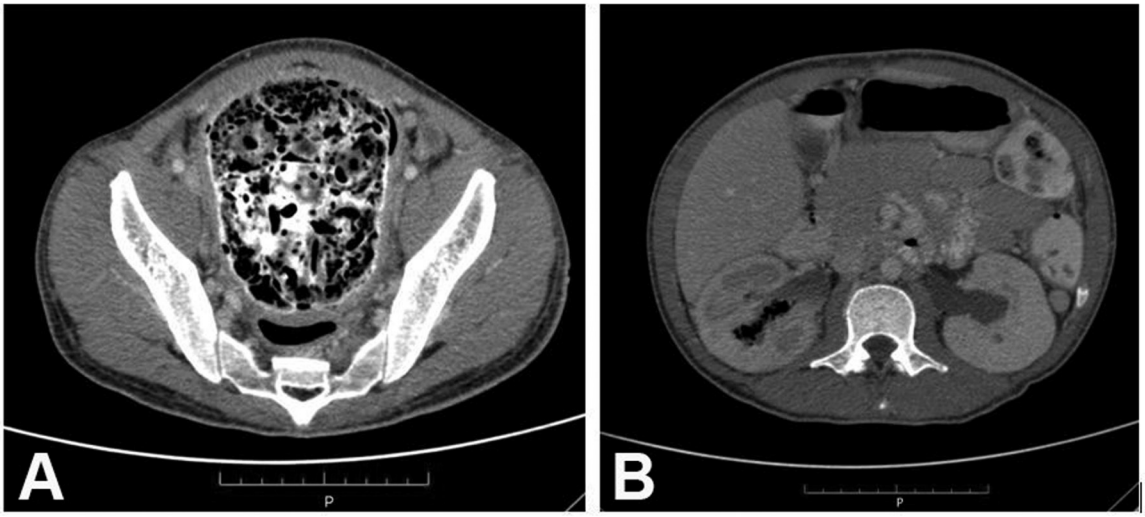

Figure 1. A) CT scan showing the urinary bladder filled with mottled soft tissue densities and air pockets; B) CT scan showing bilateral pelvocaliectasia and an emphysematous right renal pelvis.

Figure 2. A) Cystoscopy showing a large, fibrinous mass (arrow) in the urinary bladder lumen; B) Gross specimen – re-constituted after endoscopic evacuation; C) Intra-operative cystogram showing a smooth bladder outline and no demonstration of a fistula.

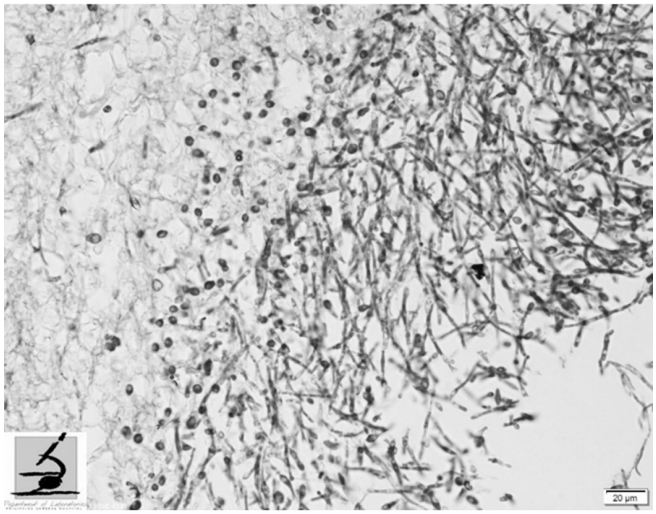

Figure 3. Histopathology slide showing hyphal elements with small yeast cells (*Candida spp*.)

Discussion

This report presents an unusual presentation of a large urinary bladder fungal bezoar in a young, undiagnosed diabetic, male who was initially managed as having an enterovesical fistula. This was because pneumaturia and passage of debris per urethra while voiding were symptoms usually associated with an enterovesical fistula.[3] Laboratory examinations eventually revealed that this patient was suffering instead from obstructive uropathy due to a large urinary bladder fungal bezoar. A possible risk factor for this condition was the uncontrolled, early-onset, type 2 diabetes mellitus of the patient. To the authors' knowledge, there are no other existing reports describing a urinary bladder fungal bezoar that presented with pneumaturia in a young male.

Fungal bezoars are commonly caused by Candida species. A bezoar, especially one that occurs within the urinary tract, is a rare condition.[1] In the last few decades, only a handful of cases have been reported. A vast majority of these cases involved the renal pelvis or ureters.[1] Fungal bezoars are usually associated with pre-mature pediatric patients who have significant anatomic or physiologic anomalies or in elderly patients who are in immunocompromised states. Examples of these are those with diabetes mellitus, malignancies, prolonged hospital stays and long-term indwelling urethral catheters.[1,2] The only risk factor present in the patient was the undiagnosed, uncontrolled diabetes.

In the few reported cases of fungal bezoar of the bladder, patients primarily presented with signs and symptoms of urosepsis and obstructive uropathy highlighted by fever, flank pain and azotemia.[1,2] None of these signs or symptoms were exhibited by the patient. Instead, the patient presented with pneumaturia, lower urinary tract symptoms and passage of debris via the urethra, all of which are uncommon presentations of a fungal bezoar.[4] Such symptoms are usually associated instead with an enterovisical fistula. It is postulated that glucosuria due to diabetes mellitus provides a urine environment that is well-suited for fungal growth. As the fungus grows, its metabolism of nutrients produces lactic acid and carbon dioxide gas as byproducts.[5] The carbon dioxide production can explain both the

pneumaturia as experienced by the patient and the air pockets on CT imaging.

The obstructive uropathy seen in the present case did not result in fungemia which could have led to sepsis. In most of the literature about fungal balls in the urinary tract, sepsis was the initial presentation.[2] Due to the high mortality of sepsis, reported to be 20-40%, early diagnosis and expedient management important.[6,7] Eliminating the cause of the obstruction or by-passing it with some form of urinary diversion is often necessary.[8] Systemic anti-fungal therapy is a cornerstone of management of fungal bezoars. In this patient, Fluconazole was highly effective in treating the fungal infection. This is mainly because it possible to achieve a higher concentration of Fluconazole in the urine compared to other anti-fungal medications.[9] It is fortunate that the management of this case was straightforward after the diagnostic dilemma had been settled.

This report on a case of a fungal bezoar in the urinary tract will help increase awareness about this rare clinical entity. Such information should help overcome the challenge of diagnosing a fungal bezoar early so that proper management can be instituted.

References

1. Takemura K, Takazawa R, Kohno Y, Yoshida S, Kato H, Tsujii T. Vesical fungus balls (fungal bezoars) by Candida albicans mimicking urothelial carcinoma in a patient with diabetic neurogenic bladder. Urol Case Rep 2018; 18: 50–1.
2. Jonczyk P, Szczerba K, Kandefer B, Potempa M, Tynior W, Kajdaniuk D. Rare cases of bezoars in the urinary tract of diabetic patients — a review of case reports. Clin Diabetol 2016; 5(4): 131–7.
3. Rovner ES. Urinary tract fistulae. In: AJ Wein (editor): Campbell-Walsh Urology. Eleventh ed. Elsevier Saunders; Philadelphia, PA: 2016; 2223–6.
4. Sobel JD, Fisher JF, Kauffman CA, et al. Candida urinary tract infections-epidemiology. Clin Infect Dis 2011; 52: S433–6.
5. McDonald DF, Fagan CJ. Fungus balls in the urinary bladder. Am J Roentgenol 1972; 114(4): 753–7.
6. Kalra O. Approach to a patient with urosepsis. J Global Infect Dis 2009; 1(1): 57.
7. Dreger NM, Degener S, Ahmad-Nejad P, Wöbker G, Roth S. Urosepsis—Etiology, diagnosis and treatment. Dtsch Arztebl Int 2015; 112(49): 837-47.
8. Davis NF, Smyth LG, Mulcahy E, Scanlon T, Casserly L, Flood HD. Ureteric obstruction due to fungus-ball in a chronically-immunosuppressed patient. Canadian Urol Assoc J 2013; 7(5-6): 355.
9. Allen D, Wilson D, Drew R and Perfect J. Azole antifungals: 35 years of invasive fungal infection management. Expert Review of Anti-infective Therapy 2015; 13:6, 787-98.