Fungal Bezoars Mimicking an Enterovesica Fistula: A Unique Case Report

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Abstract
Fungal colonization or infection of the urinary tract system is relatively common in patients with diabetes or a compromised immune system. However, fungal intravesical bezoars are extremely rare. We present a unique case with multiple, gas-holding fungals bezoars and emphysematous cystitis caused by \textit{Candida tropicalis}.

Introduction
Fungal colonization or infection of the urinary tract system is relatively common in patients with diabetes or a compromised immune system. Fungal intravesical bezoars however are extremely rare. We present a unique case with multiple, gas-holding fungal bezoars and emphysematous cystitis caused by \textit{Candida tropicalis}.

Case Report
A 74-year-old male patient with a history of type 2 diabetes mellitus, chronic kidney failure, atrial fibrillation was referred to the emergency department because of macroscopic hematuria, pneumaturia and myctalgia since a few weeks, for which the general practitioner already had started 3 courses of antibiotics.

Laboratories showed no leukocytosis, creatinine of 1.46 mg/dl, eGFR of 49 ml/min/1.73 m\textsuperscript{2} and a hemoglobin of 8.9 g/dl. Urinalysis showed pyuria and massive hematuria. A treatment with fluconazole and amoxycillin with clavulanic acid was empirically initiated.

Ultrasound demonstrated multiple intravesical, hyperechoic semicircular structures with coarse acoustic shadowing, mobile under external pressure. The scout-view showed round densities mingled with air bubbles, packed together in the pelvis. These lesions were ball-shaped and had a mottled aspect on computed tomography (CT), resembling intravesical feces. Average density measurement in these lesions were only slightly higher to large bowel stool (mean 96.6 Hounsfield units) (fig.1).

Urine culture showed a \textit{Candida tropicalis} and fluconazole 200 mg daily orally was continued. After 3 days of systematic antifungal therapy a cystoscopy was performed which showed large, white balls very suggestive for fungal bezoars (fig.2). The fungal bezoars were endoscopically resected and rinsed way under general anesthesia after an additional week of orally fluconazole 200 mg daily.
Continuous rinsing with saline was enhanced during the next 2 days after which the catheter was removed and the patient was discharged. An additional daily dose of 100 mg orally fluconazole days was prescribed for the next 14 days.

Microscopic examination of the fragments evacuated by bladder wash showed numerous budding yeast and nonbranching pseudohyphae. The yeastlike forms are oval end measure 3–5 µm and join end to end to the pseudohyphae. These morphological features are consistent with Candida species.

The patient presented at the follow-up consultation 14 days after surgery and was satisfied. He complained no more of haematuria or pneumaturia. Ultrasound showed no residual, intravesical debris or air. Urine culture showed < 10,000 Candida tropicalis.

Discussion

An increase of 300% in the prevalence of opportunistic fungal infections over the last decade has been described with Candida albicans as most common fungus [1, 2]. Although candiduria is mostly asymptomatic and requires no further treatment, underlying immunosuppression can lead to candidemia which carries a significant morbidity and mortality rate [3]. Patients with diabetes are particularly susceptible to these opportunistic infections because of their immunodeficiency but also due to glucosuria which is an excellent growth medium for the fungi [4]. Other predisposing factors are recent broad-spectrum antibiotic usage, indwelling catheters and urinary tract instrumentation [5].

Although candiduria is relatively common and is yielded in 5% of the urinary cultures and even in 26.5% of the urinary tract infections with indwelling catheters [3], intravesical bezoars are extremely rare and only 20 cases have been reported since their first description in 1961 [6]. Fungal bezoars with emphysematous cystitis are even more rare and are only described 3 times [7]. This is to our knowledge the first description of several, large, loose intravesical fungal bezoars with a faeces-like appearance on CT-scan.

The treatment of fungal bezoars is still controversial and the Infectious Disease Guidelines were based on low-quality evidence. An operative and a non-operative approach could be used. A literature study was performed by Rohloff et al. [2] in which 15 case reports were compared. They concluded that both options had advantageous outcomes. The surgical approach consists of endoscopic bladder wash, transurethral resection of the bezoars and cystotomy. The medical approach involves local antifungals, spontaneous expulsion or systemic antifungals.

Fig. 1. Axial reconstruction of an abdominal computed tomography scan. The bladder is filled with fluid and fecaloid-like material. A small amount of gas is seen in the anterior part of the bladder. The fecaloid material in the bladder is a fungus producing gas due to anaerobic metabolism.

Fig. 2. Endoscopic removal of the fungal bezoars.
We performed a surgical washout of the loose fungal balls after 10 days of systemic antifungal therapy, continuous rinsing for 48 hours with saline post-operatively and 14 days of adjuvant fluconazole. The use of amphotericin B bladder irrigation (ABBI) is controversial and expensive due to the lack of reimbursement. To our knowledge, no randomized trials compare saline rinsing to ABBI for fungal bezoars. Tuon et al. [8] performed a meta-analysis comparing ABBI with systematic antifungals in patients with asymptomatic therapy and showed that ABBI gives better *Candida* clearance rates in the first day but no difference was observed after 1 week. ABBI could be preferred in patients with renal insufficiency with an optimal treatment duration of 5 days, but it should only be used for asymptomatic candiduria because it doesn’t offer systemic therapy according to the authors. Jacobs et al. [9] report in their randomized controlled trial in elderly patients with fungal urinary tract infection a higher eradication rate in patients treated with ABBI but a higher mortality rate was observed compared to patients treated with systemic antifungals.

Sobel et al. [10] performed a randomized, controlled trial concerning fungal eradication using fluconazole or placebo in patients with asymptomatic candiduria. The clearance rate in patients with a bladder catheter was similar in treated and untreated patients. They conclude that antifungal therapy may only result in a short-term clearance and that catheter removal is the main treatment to prevent further colonization.

Surgical and non-surgical therapies are as such comparable and the potential for higher clearance rates in surgical therapy should be weighed against the operative risks.

**Conclusion**

Fungal bezoars and emphysematous cystitis should be suspected in patients with diabetes mellitus or immunosuppression complaining of pneumaturia. Treatment options include surgical and non-surgical methods in which the higher clearance rate by surgery should be weighed...