Urinary bladder perforation due to encrusted cystitis: A rare entity

Faouzi Mallat, Wissem Hmida, Khaled Ben Ahmed, Sarra Mestiri, Faouzi Mosbah

ABSTRACT

Introduction: Encrusted cystitis is extremely rare and severe chronic infection of the bladder causing intolerable function and serious consequences for the patient.

Case Report: Herein, we report a new case of encrusted cystitis complicated by rupture of the bladder, septic peritonitis and uroperitoneum, and digestive fistula in a 57-year-old male and review literature.

Conclusion: Encrusted cystitis is a severe entity that its evolution is unpredictable and it may be complicated by urinary bladder rupture that should be considered.
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Keywords: Encrusted cystitis, Bladder rupture, Septic peritonitis, Uroperitoneum, Digestive fistula

INTRODUCTION

Encrusted cystitis is a severe chronic infection of the bladder. It is a very rare form of chronic cystitis [1]. It is characterized from a functional perspective by the usual symptoms of cystitis and inconstantly by the elimination of encrusted crystals and even stones during micturition. Calcified plaques composed of calcium salts including phosphate, carbonate, and ammonium-magnesium salts in the bladder mucosa accompanied by inflammation and ulcerations characterize pathologically encrusted cystitis. Despite its increasing incidence in nineties, especially in immunodepressed patients and renal transplant recipients, less than 100 patients have been reported in the published articles, mainly single case reports. Etiology of this rare entity is presently controversial. A few hypotheses exist to explain this phenomenon. The most popular of them proposes combined action of mucosal alterations and microorganisms that split urea forming ammonia and thus creating an alkaline environment leading to the disease by precipitation of calcium salts. Corynebacterium group D2 was denoted as the most frequent culprits [2]. Therapeutic strategies are not well-defined owing to the rare occurrence of this entity.

No cases of urinary bladder rupture due to encrusted cystitis have previously been reported to our knowledge.

CASE REPORT

A case of a 57-year-old male without pathologic past history was presented with a peritonitis secondary to rupture of an encrusting and necrotizing cystitis. He had experienced dysuria, suprapubic pain, intermittent macroscopic hematuria, elimination of stones, anorexia and weight loss of about 12 kg over the previous four months.

Our patient initially presented to the accident and emergency department early in the morning with a sudden onset of sharp and constant lower abdominal pain, mostly located in the hypogastrium. The pain became worse and was associated with symptoms of oliguria and dribbling. On examination, there was generalized tenderness,
guarding and rigidity in the lower abdomen with positive rebound tenderness.

All the initial blood results were normal but the white blood cell count was $32 \times 10^9/L$. Urinalysis revealed a pH of 8.5 and was positive for the presence of blood and leukocyte esterase.

Abdominal radiograph was inconclusive. Sonogram of bladder revealed thickening of bladder wall with two distinct layers. Superficial layer (white arrow) is echogenic, corresponding to encrustation of urothelium. Underlining layer (black arrow) is hypoechogenic, corresponding to detrusor.

Computed tomography (CT) scan showed free fluid in the abdomen with a thickening of a limited segment of the small bowel, thick calcification of urothelial wall, not mobile in prone position (Figure 1) and moderate bilateral hydronephrosis.

Based on the clinical and radiological findings, the patient underwent laparotomy.

Exploratory laparotomy showed the cause of the patient’s symptoms—intraperitoneal rupture of the urinary bladder. We revealed an area of focal necrosis, thickening and calcification of the bladder wall leading to peritonitis and uroperitoneum (Figure 2). The lesion was found to be completely adhered to the bladder wall making resection difficult. It had necrotic edges and a petrous consistency when cut. A fistula between the bladder and the sigmoid has been identified (Figure 3). Partial resection of the lesion was carried out. The defect in the wall of the urinary bladder was sutured and a colostomy was associated there.

Definitive histopathological study reported calcic crystal deposit with data of non-specific chronic cystitis resulting in diagnosis of encrusting and necrotising myositis (Figures 4–6).

In postoperative period, the patient was managed with third generation cephalosporins and aminoglycoside. Corynebacterium (group D2) was present in the specific culture.

The evolution was rapidly fatal on the fifth postoperative day by septic shock.

**DISCUSSION**

Encrusted cystitis is a rare and severe inflammation of the bladder mucosa [1, 2]. It was described first in 1914 as a more or less localized ulcerated inflammation of the
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bladder wall with calcium phosphate deposits on the ulcerated surface and walls [3].

The fundamental factor for its existence is a precipitated salt deposit which requires alkaline urine. Currently Corynebacterium urealyticum is almost exclusively described in this disease [4]. Corynebacterium urealyticum is a gram-positive commensal microorganism of the skin [5]. Urinary infections due to this bacterium require three conditions to cause alkaline-encrusted cystitis: a clinical context with immunosuppression or prolonged antibiotic therapy; urologic procedures either surgical or endoscopic, and an inflammatory or neoplastic pre-existing lesion of the urothelium [5, 6]. The delay between the urologic procedure and the diagnosis can vary up to several years [5]. It was demonstrated that its active osteogenic process is reversible, probably by changing the tissue’s environmental conditions (reducing inflammation and eradicating infection), suggesting that Corynebacterium urealyticum infection can be considered the ‘primum movens’ of pathogenic mechanisms behind the bladder tissue calcification [4].

In some reported cases, this bacterium was not found because it was not specifically sought [7]. Our patient had all the clinical and histological features of Corynebacterium urealyticum infection.

Mitomycin C as a bladder chemotherapeutic agent can be a factor in the development of the disease [8].

On cystoscopy, encrusted cystitis is characterized by a marked inflammatory appearance of all or part of the bladder mucosa with ulcerations and whitish plaque corresponding to multiple calcified encrustations [2].

Ultrasound is sensitive for encrusted cystitis diagnosis and shows thickening and calcified lesions in the bladder wall. Computed tomography scan is a more sensitive technique to detect calcification even if it is thin or radiolucent on radiographs. It provides excellent visualization of the urothelial wall and calcification. It should be considered in deciding optimal treatment and in monitoring the regression of calcified plaques [9].

Encrusted cystitis is a severe chronic infection of the bladder causing intolerable function and serious consequences for the patient. It may be revealed by some non-specific complications such as urinary tract obstruction, macroscopic hematuria, renal failure. Encrusted cystitis may be complicated by rupture of the bladder, and to the best of our knowledge no previous cases of urinary bladder rupture due to encrusted cystitis have previously been reported.

In this case, the severe chronic inflammation, important edematous mucosa, huge calcifications associated with obstructive urinary problems long neglected may contribute to the fragilisation of the urinary bladder wall and its rupture.

Treatment consists of three complementary elements: treatment of infection, urine acidification and chemolysis, and elimination of calcified plaque that contains microorganisms. All Corynebacterium group D2 strains are sensitive in vitro to glycopeptides, vancomycin, and teicoplanin and have a similar effect. Calcified plaque contains high levels of microorganisms and limits in vivo antibiotic effectiveness. Surgical or endoscopic plaque removal is suggested. Transurethral resection of bladder plaque remains difficult with risk of segment rupture.

Bladder wall healing depends on calcium precipitation prevention and therefore on the prevention of calcium salt oversaturation in alkaline urine [10].

**CONCLUSION**

Encrusted cystitis is a rare and severe chronic inflammatory disease of the bladder. Its diagnosis is difficult and should be considered when presented with a long history of symptoms associating dysuria, suprapubic pain, intermittent hematuria, persistent elimination of stones. Urinary bladder rupture as a complication and a
circumstance of discovery of the encrusted cystitis is an exceptional condition.

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Guarantor
The corresponding author is the guarantor of submission.

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Authors declare no conflict of interest.

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