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.
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.

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 with 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
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

---

**Figure 5:** Necrosis of bladder wall with presence of calcic deposits (H&E stain, x200).

**Figure 6:** Extensive necrosis of the cystic wall with diffuse hemorrhagic and polymorphous inflammatory infiltrate (H&E stain, x200).
circumstance of discovery of the encrusted cystitis is an exceptional condition.

**********

Acknowledgements
We are grateful to Professor Faouzi Mosbah for his comments on the manuscript.

Author Contributions
Faouzi Mallat – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Wissem Hmida – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Khaled Ben Ahmed – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sarra Mestiri – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Faouzi Mosbah – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

Copyright
© 2014 Faouzi Mallat et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES
1. Tanaka T, Yamashita S, Mitsuzuka K, et al. Encrusted cystitis causing postrenal failure. Journal of Infection and Chemotherapy 2013 Dec;19(6):1193–5.
2. Jelic TM, Roque R, Yasar U, et al. Calcifying nanoparticles associated encrusted urinary bladder cystitis. International Journal of Nanomedicine 2008;3(3):385–90.
3. Francois J. La cystite incrustée. J Urol MA Chir 1914;5:35.
4. Del Prete D, Polverino B, Ceol M, et al. Encrusted cystitis by Corynebacterium urealyticum: A case report with novel insights into bladder lesions. Nephrol Dial Transplant 2008;23(8):2685–7.
5. Meria P, Desgrippes A, Arfi C, Le Duc A. Encrusted cystitis and pyelitis. J Urol 1998;160(1):3–9.
6. Coyle MB, Lipsky BA. Coryneform bacteria in infectious diseases: clinical and laboratory aspects. Clin Microbiol Rev 1990;3(3):227–46.
7. Agudo JM, Morales JM, Salto E, et al. Encrusted pyelitis and cystitis by Corynebacterium urealyticum (CDC Group D2): a new and threatening complication following renal transplant. Transplantation 1993;56(3):617–22.
8. Pascual Regueiro D, García Sánchez S, Oliva Encina J, Remón Garijo ML, Martínez Begochea J, Abril Baquero G. Incrusted cystitis after Mitomycin-C. Actas Urol Esp 2005 Jul-Aug;29(7):715–8. [Article in Spanish].
9. Thoumas D, Darmallaicq C, Pfister C, et al. Imaging characteristics of alkaline-encrusted cystitis and pyelitis. AJR Am J Roentgenol 2002;178(2):389–2.
10. Castellanos-Hernández H, Landa-Soler M, Venegas-Ocampo P, et al. Encrusting cystitis secondary to intravesical chemotherapy with mitomycin C. Rev Mex Urol 2011;71(1):31–5.
Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals
Edorium Journals is a publisher of high-quality, open access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission
We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?
In less than 10 words - we give you what no one does.

Vision of being the best
We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services
We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review
All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review
All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version
Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status
From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks
You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.

Four weeks
After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.

Mentored Review Articles (MRA)
Our academic program “Mentored Review Article” (MRA) gives you a unique opportunity to publish papers under mentorship of international faculty. These articles are published free of charges.

Favored Author program
One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program
Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence
We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...
We request you to have a look at our website to know more about us and our services.

We welcome you to interact with us, share with us, join us and of course publish with us.

CONNECT WITH US

Edorium Journals: On Web
Browse Journals

This page is not a part of the published article. This page is an introduction to Edorium Journals and the publication services.