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The importance of surgical treatment in encrusted cystitis and pyelitis: A case report

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ABSTRACT

INTRODUCTION: Encrusted cystitis and Encrusted pyelitis are rare chronic inflammatory diseases. Those conditions are commonly caused by the *Corynebacterium spp*. especially the *type D2* which is a gram positive, aerobic, slow-growing, and urea-sliting bacteria with a multi-antibiotic resistant profile.
PRESENTATION OF CASE: We report the case of a 62-year-old man with a past history of chronic obstructive pulmonary disease. He was referred to the department of urology for urosepsis. Bacterial culture results were positive to *Corynebacterium* urealyticum. The diagnosis of encrusted cystitis and pyelitis were highly considered. An adapted antibiotherapy was undertaken using vancomycin during 3 weeks. The patient presented two acute peritonitis : the first was caused by a spontaneous bladder dome rupture which was surgically repaired and the second was caused by a total bladder rupture which required cysto-prostatectomy and bilateral ureterostomy. The post operative outcomes were uneventful. Bacterial urinalysis was negative and total recovery was obtained.
DISCUSSION: In the majority of the reported cases, there were no sepsis or peritonitis conditions. Medical treatment by the glycopeptides and urine acidification was sufficient. However in this case, the sepsis condition and the bladder rupture with acute peritonitis made exclusively medical treatment by antibiotics insufficient. Therefore cystectomy associated to conventional antibiotics were able to limit the systemic dissemination of the bacteria and save the patient's life.
CONCLUSION: Glycopeptides antibiotics are currently the preferential treatment of encrusted cystitis. In some complicated conditions such as bladder rupture and urosepsis as in this case, radical surgical treatment by cystectomy must be realized early to avoid peritonea and septic shock.

1. Introduction

Encrusted cystitis (EC) and encrusted pyelitis (EP) are rare chronic inflammatory diseases [1].

Those conditions are commonly caused by the *Corynebacterium spp*. especially the *Corynebacterium type D2 (CBGD2)* which is a gram positive, aerobic, slow-growing, and urea-sliting bacteria with a multi-antibiotic resistant profile [2].

The first description of the EC was made in 1914 by François J [3]. In fact, few cases have been reported in the literature.

The usual clinical presentation is some non specific urinary symptoms including dysuria, pelvic pain, hematuria and rarely voiding mucus or calcified mucopurulent stones [2,4].

Diagnosis and optimal treatment have not been well established yet.

We report the case of an EC associated with an EP in an adult male.

2. Presentation of case

A 62-year-old man with a past history of chronic obstructive pulmonary disease and recent ischemic cerebral stroke was referred to the department of Urology for an acute urosepsis. The patient was suffering from recurrent cystitis and a spontaneous rupture of the right upper urinary tract treated by antibiotics only (Ciprofloxacin).

On admission, the patient presented fever, pelvic pain, dysuria and urinary retention which was treated by a transurethral catheter.

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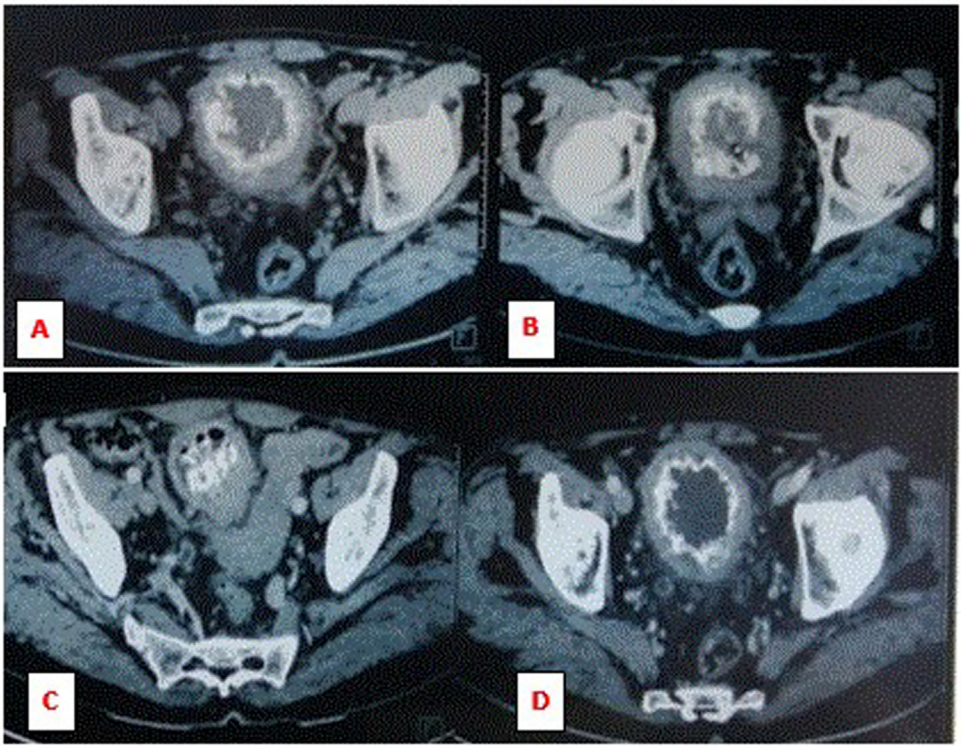

Fig. 1. Cross sectional pelvic CT Scan images (A + B: Unenhanced CT scan and C + D: Enhanced CT scan) showing thickening of bladder wall with thick and irregular encrusted calcifications.

On examination, the patient was febrile (39°c) with tachycardia and low blood pressure (90/50 mmHg). Digital rectal exam was painful and abdominal examination was normal.

Blood analysis showed a neutrophilic leukocytosis and a high C-reactive protein level (256 mg/L).

Urinalysis revealed alkaline urine (ph = 7,6) with pyuria and hematuria.

The diagnosis of an urosepsis caused by urinary infection was highly suspected.

The patient was hospitalized, probabilistic antibiotherapy based on ofloxocin and ceftriaxone was prescribed until urinary culture results are ready.

A CT scan was performed and showed a calcified thickening of the whole bladder (Fig. 1), a moderate hydro-nephrosis in the right side with a small calcification on the right renal pelvis. (Fig. 2).

After 24 h, the patient was afebrile, without tachycardia and blood pressure was normal.

Regarding this radiological aspect of the bladder, a cystoscopy was performed seven days later and showed a calcified, hyperemic and fragile bladder with a thickened wall involving the whole bladder (Fig. 3).

Multiple biopsies during endoscopy were performed.

Many diagnoses were suspected such as muscle invasive tumor, urinary tract bilharziasis, tuberculosis and encrusted cystis.

Biopsies were sent for pathological and parasitological analysis.

In addition, a non-specific and specific bacteriological culture for *Corynebacterium* and tuberculosis were carried out.

Bacterial culture results were positive to *CBGD2*.

The diagnosis of encrusted cystitis and pyelitis was highly considered.

An adapted antibiotherapy was undertaken using vancomycin 15 mg/kg every 8 h during 3 weeks.

Four days later, the patient presented an acute peritonitis with acute abdominal pain and abdominal tenderness.

A laparotomy was performed urgently. An unusual Bladder rupture in the dome, far from biopsy location, was found. Surgical repair was realized.

On pathological examination, the photomicrograph showed an extensive bladder mucosal ulceration with necrosis and microcalcifications and increased vascularity of the lamina propria (Hematoxylin and Eosin (HE) ×40) (Fig. 4A). A high power view showing microcalcifications and necrotic debris surrounded by an inflammatory cellular infiltrates of lymphocytes, eosinophils, histiocytes and foreign body giant cells (HE ×400) (Fig. 4B).

The diagnosis of encrusted cystitis was confirmed by histological findings.

A cystectomy was discussed but we opted for a conservative treatment based on literature finding.

The diagnosis of encrusted cystis was confirmed by anatomopathological exam.

Seven days later, a second laparotomy was performed because of an acute peritonitis due to complete bladder rupture. The Bladder reparation was not possible and a complete cysto-prostatectomy and bilateral ureterostomy were performed.

The post operative outcomes were uneventful. Vancomycin was carried on during 3 weeks. Bacterial urinalysis was negative.

Control CT-scan showed pyelic calcification regression and the patient was discharged home.

3. Discussion

Encrusted cystitis is a rare chronic inflammatory disease of the bladder.

The major etiological agent of encrusted cystitis and pyelitis is the *CBGD2*.

This microorganism is a commensal bacterium of the skin present in 12% of the population [5]. Patients with past history of intravesical chemotherapy or BCG installations or urinary trauma may have a higher risk to get an Encrusted cystitis [6].

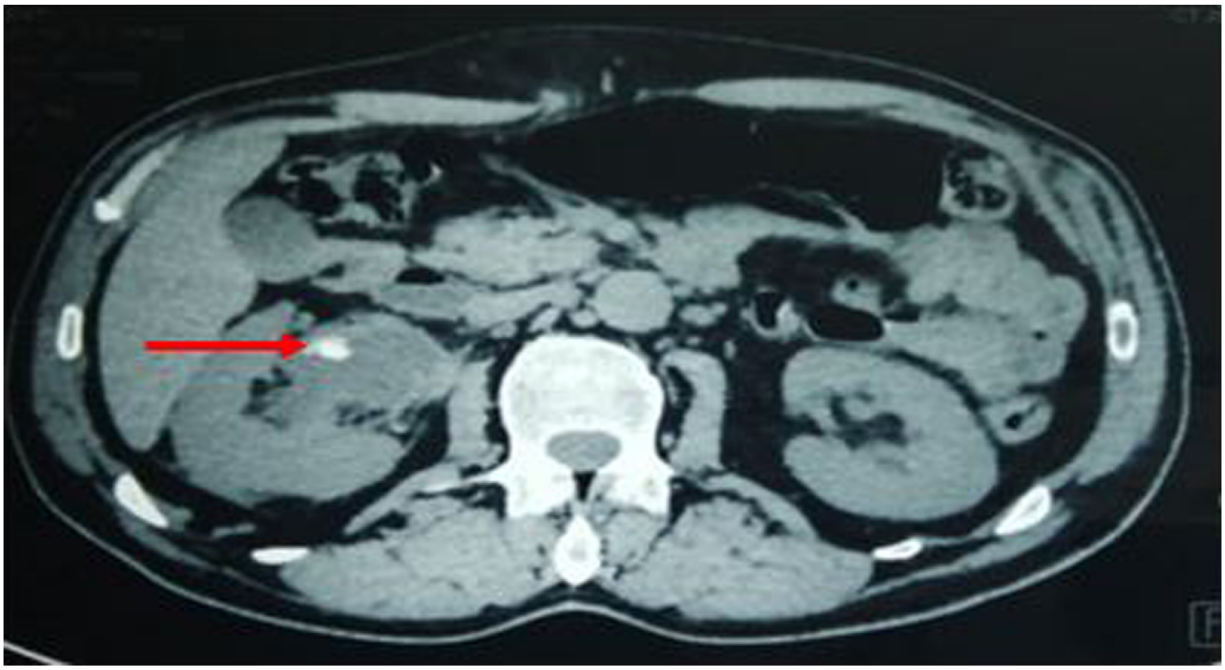

Fig. 2. Cross sectional abdominal unenhanced CT Scan image showing right hydronephroses with thickening of therightpyelo-uretheral wall and superficial pyelic encrusted calcification (red arrow).

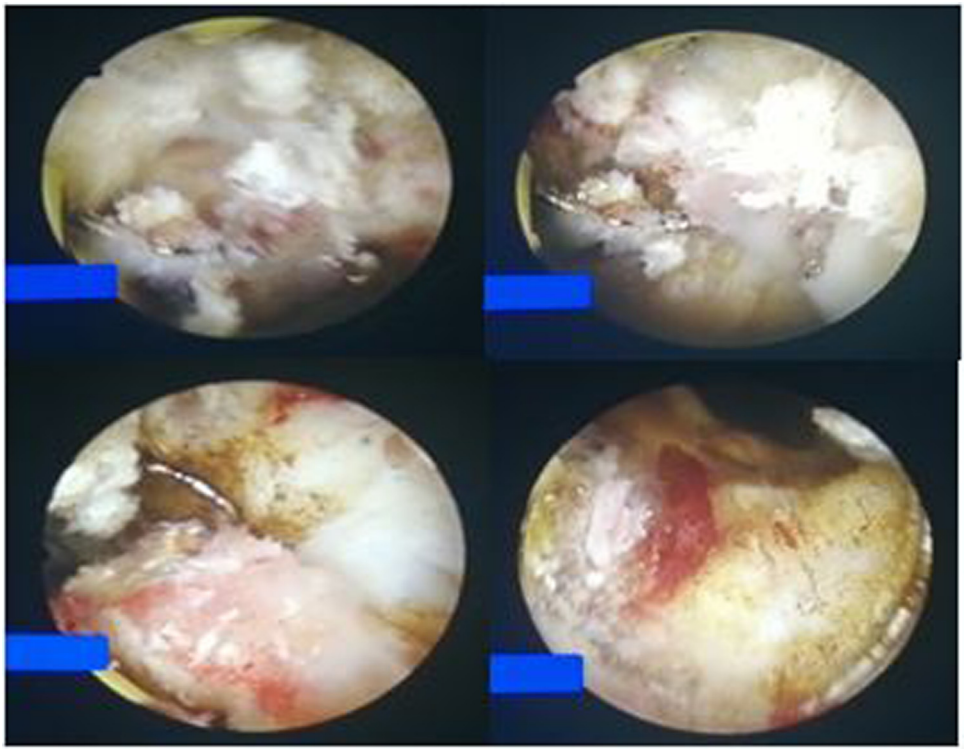

Fig. 3. Endoscopic imaging showing a large calcified and hyperemic thickening involving the whole bladder wall.

Corynebacterium urealyticum have a slow growing bacterial urease activity resulting in ammonia release, which damages the glycosaminoglycanlayer of the bladder mucosa. As a result, calcified plaques in the interstitium of the bladder mucosa occur [7].

The diagnosis of encrusted cystitis or pyelitisis is often made late. This can be explained by the lack of specific signs and also by the fact that the successful isolation and culture of *C. urealyticum* requires culturing for 48–72 hours at 37 °C on enriched media with sheep blood agar or 5% carbon dioxide [3].

Indeed, having a positive urine culture for C urealyticum does not necessarily mean that the patient has calcified encrusted particles in the urothelial mucosa. EC incidence was reported as 15.6% for patients with positive C urealyticum urine culture [8].

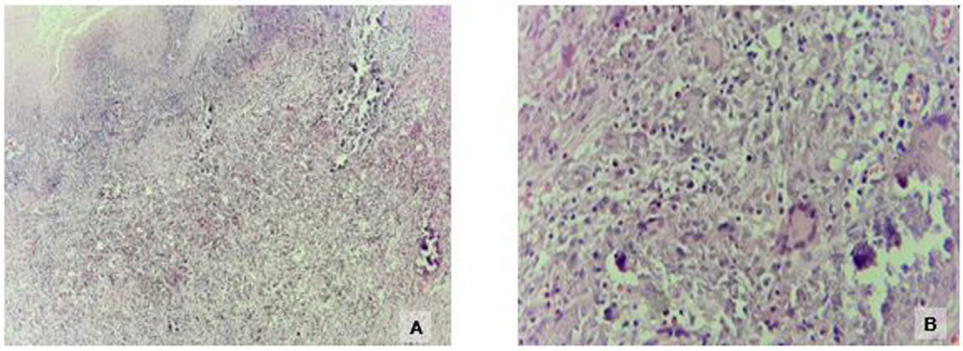

Fig. 4. (A) Photomicrograph showing extensive bladder mucosal ulceration with necrosis and microcalcifications and increased vascularity of the lamina propria (Hematoxylin and Eosin (HE) ×40). (B) High power view showing microcalcifications and necrotic debris surrounded by an inflammatory cellular infiltrates of lymphocytes, eosinophils, histiocytes and foreign body giant cells (HE ×400).

In current literature, diagnosis is generally made by finding the C urealyticum in the urine and in the pathological exam.

CT scan is a useful tool for the diagnosis, because it shows thickening and calcified lesions on the bladder's mucosa. It appears to be the best technique to diagnose encrustation particularly in the upper urinary tract [9].

Glycopeptides are the most effective antibiotics currently used as a first choice treatment of encrusted diseases.

All strains of *C. urealyticum* are susceptible in vitro to vancomycin and teicoplanin, and these drugs showed efficacy in an experimental rat model of encrusted cystitis [10].

In the majority of the reported cases, there were no sepsis conditions. Medical treatment by the glycopeptides and urine acidification was useful [4].

In this case, the sepsis condition and the bladder rupture with acute peritonitis made medical treatment exclusively by antibiotics insufficient.

Cystectomy (as a removal of encrusted lesions) and the use of conventional antibiotics were necessary in our case to limit the systemic dissemination of the bacteria and to save the patient's life.

Cystectomy as a radical surgical solution was discussed in the beginning because of the importance of bladder lesions but was not approved because of the lack of literature evidences and the high morbidity and mortality rates of this operation.

To summarize, cystectomy plus glycopeptides antibiotics should be recommended as a suitable alternative for patients with uncontrolled sepsis or a complicated cystitis like spontaneous bladder rupture.

Further studies are needed to confirm our treatment effect and results.

4. Conclusion

Encrusted cystitis and pyelitis is a serious urinary infection characterized by encrustations in the wall of the urinary tract. Diagnosis must be considered in patients with chronic urinary symptoms, alkaline urine analysis and a calcified bladder thickening on CT-scan. Glycopeptides antibiotics are currently used. In some complicated conditions such as bladder rupture and urosepsis as in our case, radical surgical treatment by cystectomy must be realized early to avoid peritonea and septic shock.

State

The work has been reported in line with the SCARE 2018 criteria [11].

Declaration of Competing Interest

The authors have no conflict of interest to declare.

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Ethical approval

Given the nature of the article, a case report, no ethical approval was required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ahmed Loghmari: Writing - original draft.
Khaireddine Bouassida: Project administration writing - review.
Oussama Belkacem: writing and editing.
Mouna Ben Othmen: review and editing.
Wissem Hmida: Supervision and reviewing.
Mehdi Jaidane: Supervision; reviewing and editing.

Registration of research studies

No registration.

Guarantor

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