

C A S E R E P O R T

Case report of life-threatening complications following cystectomy in a woman with neurogenic lower urinary tract dysfunction treated with indwelling bladder catheter for about 30 years

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Summary. Patients with neurogenic lower urinary tract dysfunction (NLUTD), specially with indwelling bladder catheter (iBC), have an increased risk of developing bladder stones, incomplete bladder emptying, recurrent urinary tract infections, sepsis, urethral trauma and bladder cancer. We present the case of a patient with a large bladder stone in iatrogenic NLUTD treated with iBC for about 30 years, who underwent a cystectomy followed by several life-threatening complications, like septic episodes and multiple surgeries. Our case outlines the importance of limiting the time of iBC in favor of CIC to avoid severe complications. Clinicians should instruct on the correct CIC technique and hygiene practices, and monitor patients with periodic abdominal US in order to diagnose and treat precociously any disease, like bladder stones. (www.actabiomedica.it)

Key words: case report, neurogenic lower urinary tract dysfunction, bladder stones, bladder catheter, intermittent catheterization, cystectomy

Introduction

Neurogenic lower urinary tract dysfunction (NLUTD) is one of the most challenging problems in urology. Various disorders or injuries affecting nervous system may cause chronic bladder dysfunction, whose type depends on central or peripheral nervous system damage level and intensity. The bladder may become overactive or underactive/acontractile. In case of underactive bladder, patients complain with voiding problems (e.g. hesitancy; straining; poor, prolonged and intermittent flow; terminal dribble), post micturition symptoms (e.g. sensation of incomplete emptying) and urinary retention. The main urodynamic finding is a reduced strength of the detrusor contraction

or its absence. Neurogenic complete or incomplete retention may require clean intermittent catheterization (CIC), which should be recommended as the first choice of treatment for its safety, especially if the patient is physically and mentally willing to perform the task or has a caregiver who is able to assist (1). Instead, indwelling bladder catheter (iBC) should be avoided because it significantly increases the risk of complications, like bladder stones, incomplete bladder emptying, recurrent urinary tract infections (UTIs), sepsis, urethral trauma and bladder cancer (2).

We present the case of a patient with a large bladder stone in iatrogenic NLUTD treated with iBC for about 30 years, who underwent a cystectomy followed by several complications.

Case presentation

A sixty-nine-year-old Italian female housewife with NLUTD was referred to our tertiary referral center because of recurrent UTIs and problems encountered during monthly iBC change. She underwent surgery for spondylolisthesis when she was fourteen-year-old. Later, she presented stress urinary incontinence treated with an increasing number of pads per die, until, at the age of 41 years, she was addressed to iBC, changed on a monthly basis by the husband. She didn't report other comorbidities and she didn't take a pharmacological therapy.

The initial diagnostic work-up consisted of abdominal cross-sectional imaging, firstly ultrasound (US), and later computed tomography (CT), showing a 41x47 mm bladder stone (Fig. 1). A videourodynamic study documented a defunctionalized bladder. She was addressed to a uterus-sparing simple cystectomy (Fig. 2) with uretero-ileal-cutaneous diversion according to Bricker, performed by a skilled surgeon, in general anesthesia. The operative time was 215 minutes. The antibiotic prophylaxis was made with Cefazolin. No intra-operative complications were recorded. Surgical pathologist identified an active chronic cystitis with follicular and eosinophilic features, associated with mucosa ulcers.

On the third post-operative day (POD), the patient was septic and complained of a severe lumbar pain. Abdominal fluid cultures revealed colonization by *Escherichia coli*, *Pseudomonas aeruginosa* and *Candida glabrata*. The CT scan documented pancre-

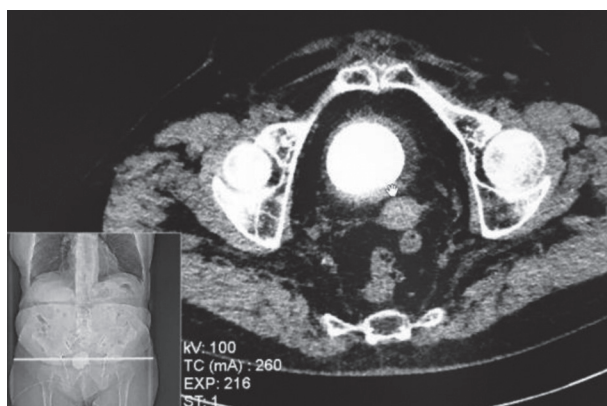


Figure 1. Abdominal CT showing bladder stone

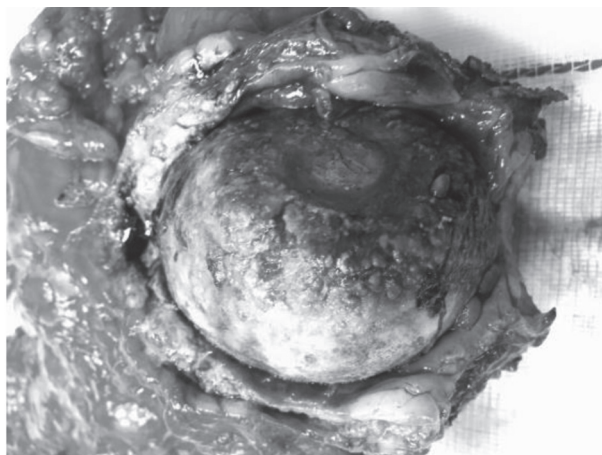


Figure 2. Sectioned bladder with inside the stone

atic oedema. A conservative approach was attempted without success. On the 18th POD, the patient had a septic shock. Blood and wound cultures were positive for, respectively, *Enterococcus faecium* + *Pseudomonas aeruginosa* and *Acinetobacter* spp. A urgent exploratory laparotomy documented acute necrotizing hemorrhagic pancreatitis. Over the next weeks, she continued medical therapy and underwent other surgeries for abdominopelvic washing. The patient had other septic episodes, prolonging her stay in intensive care (95 days in total). The patient was discharged after 141 days. The complications reported can be classified as a IV grade of Clavien-Dindo scale.

At the last follow-up visit, two years later, the patient had a well-functioning urinary diversion and the urinary tract didn't present any significant conditions.

Discussion

In this paper, we present the case of a patient with a large bladder stone in iatrogenic NLUTD treated with iBC for about 30 years, who underwent a cystectomy followed by several life-threatening complications. As a case report, this paper is associated with a low level of evidence and it describes an uncommon and extreme clinical situation for lower urinary tract lithiasis.

To our knowledge, this paper is the first report of a voluminous bladder stone connected to iBC, leading to cystectomy, which revealed being associated with several life-threatening complications.

In literature similar cases are reported, but in patients who underwent enterocystoplasty or orthotopic neobladder diversion. These patients usually perform CIC. This situation is associated with bladder stones, which may require surgical treatment, as, if they left untreated, they will continue to grow up, representing a source of chronic UTIs. The current strategy for bladder stones in patients with NLUTD is endoscopic electrohydraulic lithotripsy or Holmium:YAG laser, while open cystolithotomy is indicated for voluminous bladder stones. In these cases, cystectomy is an extremely rare condition, a last-resort treatment, usually in patients with a dysfunctional fibrotic bladder, like the case presented in this paper.

The patient underwent surgery for spondylolisthesis when she was fourteen-year-old. Neurosurgical interventions on the rachis are a risk factor for NLUTD. In these patients, the appropriate management is represented by CIC, as iBC defunctionalizes bladder, provoking fibrosis and loss of elasticity. The principle of CIC is to follow the bladder physiology to enhance storage function and to empty it efficiently avoiding complications. The chance of successful bladder catheter removal decreases with time of iBC. Therefore, the choice of iBC in a young woman was not appropriate.

iBC is associated with various complications such as UTI, urethral trauma, bleeding, urethritis, fistula, bladder neck incompetence, sphincter erosion, bladder cancer, allergy and a defunctionalization of the vesical mechanics (2). In particular, a high incidence (46-53%) of bladder stones in patients managed with long-term iBC has been reported (2-3). Ord et al. demonstrated that the absolute annual risk of stone formation in patients with a catheter is 4% compared with 0.2% for those on CIC, and, in patients treated for previous stone, the risk of forming a subsequent stone quadruples to 16% per year. Moreover, bladder stones are no more likely to form in patients with suprapubic catheters compared to those with indwelling urethral catheters (4). The increased risk of bladder stones formation occurs independently of age, sex and injury level (4).

Therefore, iBC should be used for as short a time as possible, shifting precociously to CIC. This step is usually demanding at the very beginning, requiring a significant effort to perform it appropriately, even if

the learning curve is steep. In patients with CIC, the most important prevention measures for UTIs are a valid education, a valuable patient compliance, the use of proper material and the application of a proper catheterization technique. It is mandatory to empty bladder at the end of the maneuver, as residual urine plays a major role in provoking UTI. The average frequency of CIC is 4-6 times per day; more frequent catheterizations increase the risk of cross UTIs (2-5). In patients in CIC with recurrent UTIs, it is possible to prescribe a low-dose antibiotic prophylaxis (e.g. nitrofurantoin, trimethoprim or caalexin). The results of a large randomized controlled trial made by Pickard and al. demonstrated a significant benefit for antibiotic prophylaxis in terms of reducing the frequency of UTI for people carrying out CIC; the frequency of symptomatic antibiotic-treated UTI was reduced by 48% using prophylaxis (6).

Another critical point of patient management was the follow-up. There is no consensus on the appropriate urological follow-up of individuals with NLUTD. In his review of literature, Cameron et al. recommended US of the urinary tract to detect urinary stones with good sensitivity (7). Therefore, it is mandatory to prescribe an abdomen US once a year or at least once every two years as a first step for the diagnosis of main complications, like urolithiasis, bladder tumors, upper urinary tract tumors etc. In this case, the patient did not undergo a periodical US follow-up.

According to authors, the above-mentioned negligence in patient management determined a compromised, infective general status, which resulted in a long, complicated hospital stay following cystectomy. The patients with NLUTD followed by our tertiary referral center are in CIC and undergo periodical US exam: the result of this approach is the absence of patients undergoing cystectomy, as CIC does not defunctionalize bladder, and vesical stones are diagnosed precociously and treated endoscopically in a day-hospital regimen.

Although this paper, as a case report, is associated with a low level of evidence, it outlines the importance of some good clinical practices, which are easily adoptable in the routine clinical practice. Firstly, iBC should be used for as short a time as possible. In comparison with iBC, the use of CIC reduces the risk of complica-

tions. Patients should be trained appropriately to the maneuver of CIC. The experience of a referral center may overcome the difficulties with learning curve of CIC. Patients with NLUTD should undergo both periodical out-patient visits and US examination, in order to diagnose precociously major complications like bladder and/or reno-ureteral stones, bladder cancer, bladder diverticula etc.

Finally, this paper presents the case of a huge bladder stone connected to iBC, which leads to a cystectomy, associated with life-threatening complications, outlining the pivot role of a correct management of NLUTD through periodical out-patients visits and US, besides CIC to facilitate the bladder physiology and reduce the risks of complications.

Conclusion

Our case outlines the importance of limiting the time of iBC in favor of CIC to avoid life-threatening complications. When it is not possible, clinicians should instruct on the correct CIC technique and hygiene practices, and monitor patients with periodic abdominal US in order to diagnose and treat precociously any disease, like bladder stones.

The patient was informed about this paper and she consented to the publication.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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