

Case Report

Recurrent giant neobladder calculus extrusion after bladder exstrophy repair

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Abbreviations & Acronyms

BE = bladder exstrophy
EIC = exstrophy-epispadias complex

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Introduction: Patients with bladder exstrophy subjected to reconstructive surgeries are at risk of developing urinary calculus.

Case presentation: We report the case of a 29-year-old male patient with bladder exstrophy who presented with a recurrent episode of extrusion of calculus through the neobladder and anterior abdominal wall. Calculus removal and reconstructive repair of the neobladder and abdominal wall were performed in 2010. Nine years following the procedure, the patient returned with new large neobladder calculus extrusion.

Conclusion: Recurrence of large calculus should be seen as the new paradigm for the importance of close follow-up in bladder exstrophy patients.

Key words: augmentation cystoplasty, bladder exstrophy, calculus, neobladder, vesicocutaneous fistula.

Keynote message

Prior medical history of bladder exstrophy repair increases the risk of neobladder dehiscence and calculus extrusion. This case highlights the importance of close follow-up of patients with bladder exstrophy and the need for an institutional protocol for longitudinal urological care from childhood to adulthood.

Introduction

The exstrophy-epispadias complex (EIC) presents with an anterior midline defect with variable expression including a spectrum of congenital disorders that affect the gastrointestinal and genitourinary systems and the pelvic floor.^{1–3} Its incidence is estimated at 1 in 30,000 to 1 in 50,000 live births and predominantly affects males.³ The presentation of EIC may range from the least severe being epispadias to the most severe being cloacal exstrophy. However, bladder exstrophy (BE) is the most common abnormality found in those patients.¹ Current standard-of-care treatment for patients with BE involves reconstruction of various aspects of the deformity during the neonatal period.² The overall goals of treatment are to optimize continence, preserve renal function, and improve genital esthetic and function. The most described complications are wound dehiscence, bladder prolapse, urethral outlet obstruction, and urinary calculus formation.^{2–4} Dealing with these often requires a multidisciplinary approach. Since the patients with neobladder calculus and Mitrofanoff catheterizable channel may not have urethral access to the bladder due to previous bladder-neck closure procedure, percutaneous suprapubic cystolitholapaxy, or open cystostomy may be required.

Here we present an unusual case of BE repair followed by a recurrent giant neobladder calculus extrusion.

Case report

A 29-year-old caucasian man with a medical history of BE, of poor socioeconomic status, was subjected during childhood to an augmentation ileocystoplasty with bladder neck closure and subsequent continent suprapubic antireflux Mitrofanoff appendicovesicostomy.

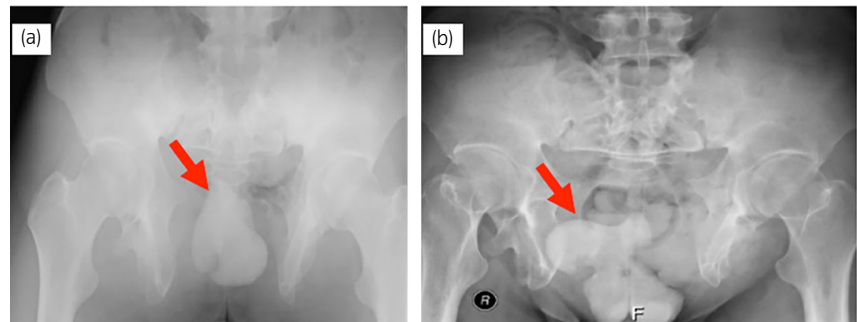


Fig. 1 Pelvic X-ray. (a) Pelvic X-ray shows an extremely large neobladder calculus (red arrow) in 2010 (b) Pelvic X-ray shows a recurrent large neobladder calculus (red arrow) in 2019.

In 2010, he developed an episode of vesicocutaneous fistula and neobladder calculus extrusion through the abdominal wall (Fig. 1a). Calculus removal and neobladder closure using a *tunica vaginalis* flap of the right testis and partial flaps of the rectus abdominis muscle was performed. Scrotal rotation skin flap was used to close the skin. There was no recurrence of the fistula nor calculus at 1 year of follow-up. The patient failed to adhere to further follow-up appointments.

In 2019, the patient developed multiple urinary tract infections, hematuria, fever, and leaking of purulent urine through the lower abdomen. Despite several visits to the hospital in his area of residence and multiple cycles of antibiotics, the vesicocutaneous fistula progressively increased in size during this period, allowing calculus extrusion to occur (Figs. 1b and 2). Four months later, the patient presented to our hospital's Emergency Department due to recurrence of calculus extrusion, fistula, and Mitrofanoff cutaneous stoma stenosis resulting in complete inhibition of self-catheterization. An incision under local anesthesia of the Mitrofanoff stoma stenosis and placement of a 10fr bladder catheter were performed to allow recanalization, with drainage of residual urine, which was collected for microbiologic analysis. Blood tests showed mild anemia, increased inflammatory biomarker levels, and mild renal dysfunction (blood urea 81 mg/dL, and serum creatinine 1.52 mg/dL). Computed tomography of the abdomen and pelvis showed a bulky neobladder calculus measuring 9.6 cm with partial extrusion and disruption of the anterior wall of the neobladder, associated with marked bilateral hydronephrosis, and kidney atrophy (Fig. 3). Piperacillin-tazobactam was initiated and he was operated on under general anesthesia. Initially, the neobladder was explored to

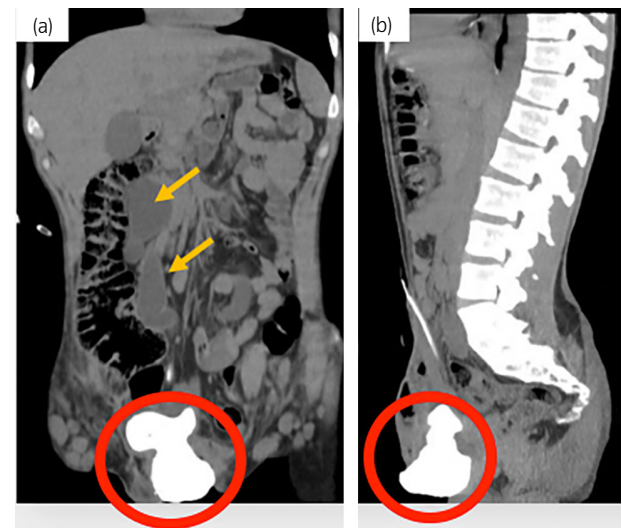


Fig. 3 Computed tomography: (a) In the coronal plane, computed tomography shows one large calculus inside the neobladder (red circle) and right kidney hydronephrosis (yellow arrow) (b) In the sagittal plane, the same large calculus (red circle) and extrusion through the abdominal wall are seen.

detach the calculus from the neobladder walls (Fig. 4). Cleaning and washing of the neobladder were performed. A 10-fr bladder catheter was replaced into the Mitrofanoff stoma, and a stoma bag was attached to the fistula. There were no complications noted at the time of procedure. Microbiologic analysis showed polymicrobial urinary culture, and the patient was discharged 10 days after the surgery.

Discussion

Bladder augmentation provides a low-pressure, continent, catheterizable reservoir for urine, preserves renal function, optimizes continence, and improves the quality of life of patients with BE. The prevalence of calculus formation reported in the neobladders varies from 3.0 to 52.5%.⁵ In general, risk factors for calculus formation include caucasian race, male sex, augmentation cystoplasty, and bladder neck repair to increase outlet resistance, urinary tract infections, foreign bodies, vesicoureteral reflux, and urinary stasis.⁵⁻⁷

Recurrence of calculi after successful removal has been reported in up to 48% of patients and may be seen as early as 3 months following removal.⁵ Patients submitted to BE

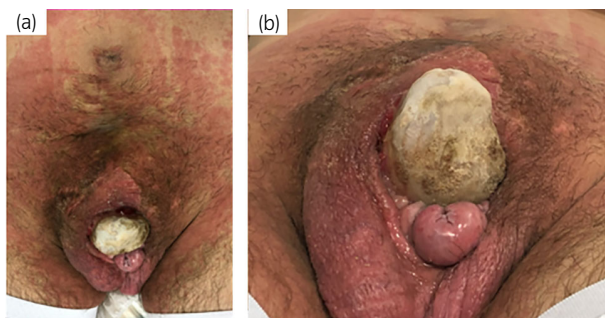


Fig. 2 Clinical findings. (a and b): Neobladder calculus extrusion through the abdominal wall.



Fig. 4 Calculus examination. (a–d) Calculus measurement.

repair are also at risk of total bladder dehiscence, bladder prolapse, neourethral stricture, and obstruction, soft-tissue loss, and vesicocutaneous fistula.^{2–4}

In both episodes, bladder outflow obstruction in a patient previously subjected to a bladder neck closure procedure and a dysfunctional and/or misused Mitrofanoff catheterizable conduit promoted urinary stasis and consequently infection and calculus formation. Further erosion of the neobladder wall by this massive calculus in a patient with a fragile lower abdominal wall due to multiples previous surgeries, added to the absence of pubic symphysis, contributed to the calculus extrusion.

We choose not to close the bladder and abdominal wall at the same time due to the risk of spreading infection and fistulae recurrence. Given the risk factors for calculus recurrence, renal function worsening, and severe infection, bladder removal, and a new urinary diversion should be considered.⁷ In this particular case, the patient was further proposed for cystectomy and construction of ileal conduit and abdominal wall and penile reconstruction.

We recommend involving the plastic surgeon in the decision-making process for adult patients with BE, especially in case of a large abdominal fistula. Different approaches for the repair of abdominal wall must be balanced with the effectiveness of the technique as well as the costs and the risks of infection and fistula, especially when a synthetic mesh is used for the repair.⁸ Attention should be given to sexual function of patients with BE since those patients can have a satisfactory sexual life.^{9,10} Grafting of the dorsal aspect of the penis should be an option. Multiple previous repairs can have a negative impact on the erectile function; hence, pharmacological strategies to improve penile erection should be considered necessary.¹⁰

In our clinical practice, since most of such patients are treated by pediatric surgery, adult urologists are less comfortable working with patients with complications of BE repair. Therefore, an institutional protocol for the transition of

urological care from childhood to adulthood is recommended.¹¹ We believe that annual imaging for assessment of the abdomen and pelvis may be a good follow-up strategy.

Conclusions

Recurrence of large calculus extrusion, which is extremely rare, should be seen as a paradigm for the importance of close follow-up and engagement of BE patients in their own care. These patients may have several complications related to bladder exstrophy repair which should be multidisciplinary managed.

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Author contributions

Thiago Guimarães: Conceptualization; data curation; validation; writing – original draft; writing – review and editing. Fátima Alves: Validation; writing – review and editing. Hugo Pinheiro: Conceptualization; validation; writing – review and editing. Luís Campos Pinheiro: Conceptualization; validation; writing – review and editing.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an Institutional Reviewer Board

Not applicable.

Informed consent

Informed consent was obtained from the patient.

Registry and the Registration No of the study/trial

Not applicable.

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