Case Report

Extensive dystrophic calcification of eroded artificial urinary sphincter cuff—a rare cause of urethral obstruction: a case report

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Background: Despite a high technical success rate and satisfaction, complications of an artificial urinary sphincter (AUS) can occasionally occur and vary in severity from transient urinary retention to unrecognized urethral injury or urethral erosion. Infection usually occurs when urine comes into contact with the device and necessitates explant of the cuff followed by delayed device replacement. In rare cases, however, the device can remain in contact with urine for long periods of time without the sequelae normally associated with acute infection.

Case Description: Here we present a case report of two patients with intraurethral migration of AUS cuffs associated with calcification resulting in urethral obstruction precluding catheterization. With extensive calcification around the cuff and longstanding complete erosion into the urethra, the urologist can expect obliteration of normal tissue planes and intense fibrosis during cuff explantation. Following excision of the cuff and stones, assessment of the urethral lumen should be performed to determine whether repair of the urethra is required. After surgery, urinary diversion with a urethral catheter is important to allow for urethral healing. Prior to considering AUS replacement, cystoscopic assessment of the urethra is critical to assess for stricture or other abnormality.

Conclusions: Extensive calcification following AUS erosion into the urethra requires the expertise of a urologist with experience in urethral surgery in order to optimize outcomes. Urologists should be aware of this uncommon, but dramatic presentation of urethral obstruction due to a chronically eroded AUS cuff.

Keywords: Artificial urinary sphincter (AUS) erosion; AUS calcification; urethral obstruction; case report

Submitted Aug 12, 2021. Accepted for publication Oct 29, 2021.
doi: 10.21037/tau-21-709
View this article at: https://dx.doi.org/10.21037/tau-21-709

Introduction

Complications from an artificial urinary sphincter (AUS) include urinary retention (31%), superficial skin infection (1%), device infection (6%), urethral atrophy (9.6–11.4%), and urethral erosion (3.8–10%) (1). If urethral atrophy goes unmanaged, the cuff may erode through the urethra. Early cuff erosion is often due to urethral injury during implantation, whereas late erosion is more often due to trauma to the cuff (1). In the absence of acute infectious symptoms, chronic erosion may result in extensive dystrophic calcification causing urethral obstruction, urinary retention, and space-filling stone debris precluding catheterization. We present the following article in accordance with the CARE reporting checklist (available at https://tau.amegroups.com/article/view/10.21037/tau-21-709/rc).

Case presentation

A 23-year-old man with spina bifida who had an AUS placed eight years prior, presented with urinary leakage around the pump, which had completely eroded through his scrotum two years before presentation. During this time, he experienced recurrent urinary incontinence, chronic urinary tract infections, and chronic bilateral epididymitis. He had been managing his bladder by cycling the cuff

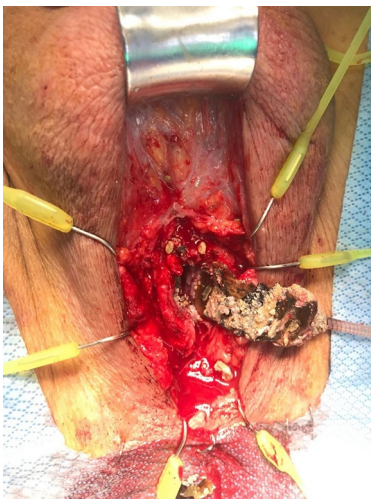

Figure 1 A perineal dissection has been carried down to the urethra. The bulbospongiosus muscle is retracted laterally with yellow hooks. The urethra has been mobilized and a vertical urethrotomy performed to extract the calcified and eroded AUS cuff. AUS, artificial urinary sphincter

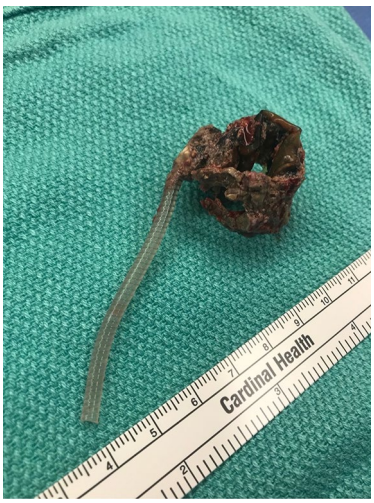

Figure 2 Explanted AUS cuff with extensive dystrophic calcification. AUS, artificial urinary sphincter

and had not required intermittent catheterizations. Upon presentation, he was afebrile and had a leukocytosis of 13,000. Physical exam demonstrated the pump completely eroded through the right hemiscrotum. After initiating intravenous antibiotics, he was taken to the operating room, at which time a urethrethroscopy demonstrated a large stone encompassing the majority of the urethral lumen.

A perineal incision was carried down to the palpable stone. A vertical urethrotomy was performed and the cuff, along with the surrounding stone, was removed. The remainder of the device components were removed and the urethrotomy was closed primarily in two layers. A #20 French urethral catheter was placed and maintained for ten days postoperatively. Due to this patient's heavily trabeculated bladder and detrusor instability, a repeat AUS was felt to have a low probability of long-term success. Eight months later, he underwent augmentation cystoplasty and formation of catheterizable appendicovesicostomy with ligation of the bladder neck.

The second patient is an 85-year-old man with SUI following radical retropubic prostatectomy. Fifteen years prior to presentation, he had an AUS placed following a failed urethral sling. Upon presentation, he remained disease free and pelvic radiation naïve. He presented with a scrotal and right groin abscess, and worsening incontinence. He had been taken to the operating room at an outside facility for an unsuccessful AUS explant due to the inflammation and calcifications around the cuff. Urethroscopy at that time reported a severely eroded urethra and stone formation on the cuff, filling almost the entire urethral lumen and preventing urethral catheter placement.

After administering intravenous antibiotics, he underwent explantation of the AUS cuff and drainage of the right groin abscess. A perineal incision was made and dissection continued to the urethra. The cuff was noted be completely within the urethral lumen. The urethra was mobilized and a dorsal midline vertical urethrotomy was performed to remove the cuff and surrounding calcification (*Figures 1*,*2*). Due to the extensive fibrotic reaction surrounding the corpus spongiosum, the prior sling was not visualized. The scarred urethra in the midline was excised. A primary closure with mucosal advancement was performed to preserve the urethral lumen diameter. A #20 French urethral catheter was maintained for ten days postoperatively. The patient is scheduled for a cystoscopy and will undergo an AUS placement eight months after device explantation. Informed consent was obtained from each patient for publication of the de-identified information presented in this case report. All procedures performed in

this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). 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 editorial office of this journal.

Discussion

AUS implantation is the gold standard treatment for SUI in men. Cuff erosion most commonly presents with acute infection, however, if patients do not develop an acute infection, the erosion may develop into a chronic process. It is not known, however, which factors predispose patients with erosion to develop a chronic process.

Chronic erosion causes urine to be in constant contact with the cuff and can result in calcification. Calcification around the cuff can become so extensive, it can result in urethral obstruction, as demonstrated by these patients. Reports of calcification of an eroded AUS device are rare, and most commonly occur in the urethra. However, this is not exclusive to the urethra, as one report describes erosion of the reservoir into the bladder in a patient who had undergone prior pelvic radiotherapy (2).

With extensive calcification around the cuff and complete erosion into the urethra, the urologist can expect obliteration of tissue planes and intense fibrosis surrounding the urethra and corpus spongiosum. After excision of the cuff and stones, assessment of the urethral lumen should be performed to determine if adjunctive maneuvers are needed to increase the urethral diameter. Urinary diversion with a urethral catheter is important to allow the urethra to heal.

Salvage continence strategies following AUS cuff erosion, including cuff repositioning and transcorporal cuff placement have been well-described (3). A continent catheterizable channel with bladder neck closure, and cystectomy with ileal conduit, are alternative options for a patient unable or unwilling to undergo another AUS placement (4). If a patient has an end-stage urethra and is not a candidate for a large abdominal operation, he may be a candidate for permanent bulbar urethral ligation (PUL) with suprapubic tube placement (5). PUL and suprapubic tube placement were not indicated in these two patients, however this may be a consideration at the time of cuff explantation in a frail man with an end-stage urethra.

Conclusions

When cuff erosion does not result in acute infection or is not recognized acutely, it can remain subclinical, and chronic erosion can present as urethral obstruction from extensive calcification on the AUS cuff. These cases require the expertise of a urologist with experience in urethral surgery in order to optimize outcomes. Urologists should be aware of this uncommon, but dramatic presentation of urinary obstruction due to chronically eroded AUS cuff.

Acknowledgments

Funding: None.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://tau.amegroups.com/article/view/10.21037/tau-21-709/rc

Peer Review File: Available at available at https://tau.amegroups.com/article/view/10.21037/tau-21-709/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tau.amegroups.com/article/view/10.21037/tau-21-709/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report, accompanying images and de-identified information. A copy of the written consent is available for review by the editorial office of this journal.

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Cite this article as: Lewis KC, Lundy SD, Angermeier K. Extensive dystrophic calcification of eroded artificial urinary sphincter cuff—a rare cause of urethral obstruction: a case report. Transl Androl Urol 2022;11(4):567-570. doi: 10.21037/tau-21-709

