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Sub-urethral Diverticulum, Including 3 Large Stones: A Case Report

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ABSTRACT

Background: Urethral diverticuli containing calculi are rare, according to the literature.
Case: A middle-aged female, with a former diagnosis of suburethral diverticulum, presented with acute pain, hematuria, and incontinence. She was found to have 3 large stones in the diverticulum from a previous MRI and was diagnosed with a urinary-tract infection (UTI) at the time of presentation. A cystourethrogram and cystoscopy was performed, and the UTI was treated. She then had an open diverticulectomy, a removal of the stones, a repair of the diverticular neck, and the placement of a Surgisis xenograft, which effectively resolved the diverticulum and stones.
Conclusion: Adequate diagnosis and treatment of urethral diverticuli containing calculi involve many components, including sufficient clinical suspicion, adequate imaging, and the prevention of postoperative complications.

INTRODUCTION

Urethral diverticuli are uncommon in women, with a prevalence of 0.6 to 6% and associated calculi accounting for 1.5 to 10% of these cases [1]. They may be asymptomatic and found incidentally, or have the following common symptoms: recurrent urinary-tract infection (UTI) (52%), dysuria (52%), urgency (41%), frequency (38%), recurrent urinary retention and a weak urinary stream (15%), stress incontinence (38%), and urge incontinence (34%) based on 32 cases. Dyspareunia and hematuria are also common findings [4,6]. Diverticuli may be complicated by stones and malignancy [2,4]. Although rare, adenocarcinoma is the most common cell type, making up 61% of malignancies [2,4].

Diverticuli are thought to be acquired from obstruction and expansion of the paraurethral skene's glands, occasionally from infection, causing weaknesses in the supportive facial layer of the bladder or urethra. Most acquired are caused by trauma from vaginal deliveries, instrumentation, and obstruction from distal urethral or meatal strictures [4,6]. A physical exam reveals a visible and palpable suburethral mass with urine or discharge frequently "milked" by palpation of the mass. Treatment often includes surgical excision of the diverticulum [4,6].

KEYWORDS: Renal arteriovenous malformation; Renal AVM; Hematuria; Ureteric obstruction
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case report

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Figure 1. Normal urethral opening on the left side of the image and the diverticular opening on the right side (patient's left side).

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What was once thought to be rare, clinical suspicion and better diagnostic imaging have aided in the detection of more diverticuli [5]. Having a patient with frequent UTIs and/or voiding problems should be an indication to further investigate urethral diverticuli.

CASE REPORT

A 54-year-old, gravida 2, para 0 female with a past medical history of total abdominal hysterectomy, bilateral salpingo-oophorectomy (TAH-BSO), and cholecystectomy presented to the gynecological-urology clinic with acute pain. She was previously seen at an out-of-state facility and was diagnosed with suburethral diverticulum with calculi but failed to have treatment. The MRI done at the time of diagnosis revealed a 2.3 cm in diameter diverticulum posterior the urethra containing 3 stones. The patient stated she had a decreased stream and blood in the urine, which was also cloudy and smelly. She stated, "something is hanging out and it hurts to do anything," and it seemed to be getting worse. In addition, the patient complained of constantly leaking urine, and had burning and pain with urination that she thought was a possible infection. She alleviated the diverticular pain by keeping her bowels soft with magnesium citrate and drank 9 to 12 glasses of water a day.

The patient had a TAH-BSO in 1995 for fibroids and endometriosis, a cholecystectomy, urinary passage of nephrolithiasis in 1998, and no other medical conditions or prior vaginal surgery. A pap smear was performed 5 months prior, with negative results, and a mammogram within the year. Her family history is positive in the father for diabetes and hypertension; her maternal grandmother with intestinal cancer, diabetes, and hypertension; and her aunts with Lupus.

The patient was married and smoked a pack over 3 days, formally a 2-pack-per-day smoker, and denied any drug use. She occasionally had wine and caffeine, but avoided soda. Her symptoms continued to keep her from her work and interfered with sexual activity. She was allergic to aspirin, Demerol, codeine, and IV contrast, and was only taking ibuprofen and Tylenol.

On exam, her blood pressure was 152/88, and she was alert and oriented although in significant distress sitting in a tilted position with the left leg raised. The exam was unremarkable, including a soft, non-tender abdomen with normal external genitalia. A suburethral diverticulum was noted on bimanual examination, along with palpable loculations and calculi. Purulent material was obtained through squeezing the mass. Urine was collected by straight catheter and sent for culture cytology.

It was decided to begin empirical treatment of a UTI with Levaquin for 21 days, followed by a cystoscopy and a urethral diverticulogram to further evaluate the size of the diverticulum opening and urethra for surgical planning. The Foley catheter was left in place for drainage. Urine results were later positive for E. coli sensitive to Macrobid, which was begun then.

A cystourethrogram showed the diverticular opening 1.5 cm from the urethrovesicle junction. Cystoscopy revealed a left-sided, 5 mm opening with urethral mucosa entering the diverticulum and no bladder abnormalities. A diverticulectomy and reconstruction was scheduled after approximately 3 weeks of antibiotic treatment and drainage of the diverticulum. A mid-urethral incision was made over the diverticulum followed by a diverticulectomy, the removal of stones (Figure 2), layered repair of the diverticular neck, a cystoscopy to evaluate any

leaking urine, and the placement of a Surgisis xenograft during closure of the vaginal mucosa. Pathology confirmed a diverticular sac without malignancy.

Figure 1 shows the normal urethral opening on the left side of the image and the diverticular opening on the right side (patient's left side).

A voiding cystourethrogram shows the 3 large stones outside the pelvis, also seen on the flat-plate X-ray (Figure 3).

The patient returned 13 days later due to irritation of the Foley catheter, which was removed only a day earlier than the intended 14-day postoperative plan. With the exception of minimal episodes of urgency a day, the patient is otherwise doing well.

COMMENTS

Urethral diverticuli are rare in women and associated calculi are uncommon, accounting for 1.5 to 10% of these cases [1].

Most diverticuli acquired are caused by trauma from vaginal deliveries, instrumentation, and obstruction from distal-urethral or meatal strictures due to infection, resulting in expansion of the paraurethral skene's glands. Our patient later gave us the most important piece of information: Over 10 years prior to her presentation, she had passed a kidney stone with much distress and discomfort.

There are multiple case reports of calculi in urethral diverticuli, although they vary in number and location. Adequate diagnosis and treatment involve many components, including sufficient clinical suspicion, adequate imaging, and prevention of post-operative complications. Like most urethral diverticuli, it appears our case was acquired from trauma. Her history of nephrolithiasis is likely the reason from possible entrapment during kidney-stone passage in the urine, thus predisposing her to diverticulum formation.

Similar to our case, a 75-year-old woman presented with dysuria and gross hematuria, and had a past medical history of UTIs and a cholecystectomy. This case was discovered by incidental finding from a routine CT scan after a motor-vehicle accident. A kidney, ureter, and bladder (KUB) X-ray was performed along with a cystourethroscopy, which was difficult secondary to a dilated

Figure 2. A mid-urethral incision was made over the diverticulum followed by a diverticulectomy and removal of stones.
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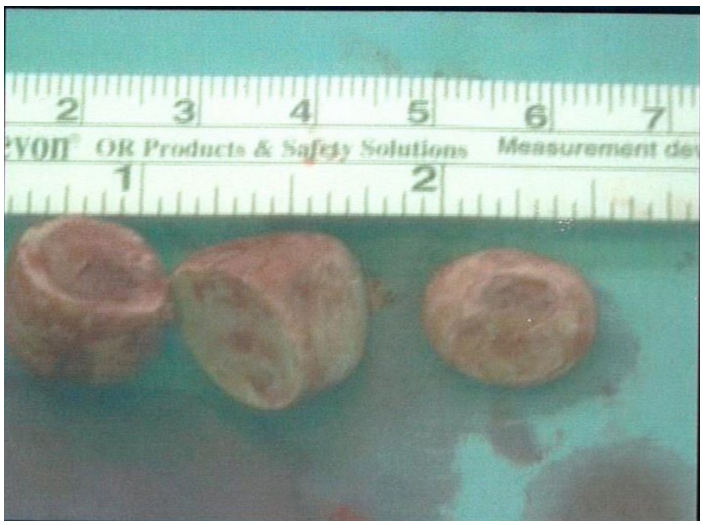

Figure 3. A voiding cystourethrogram shows the 3 large stones outside the pelvis, also seen on the flat-plate X-ray.
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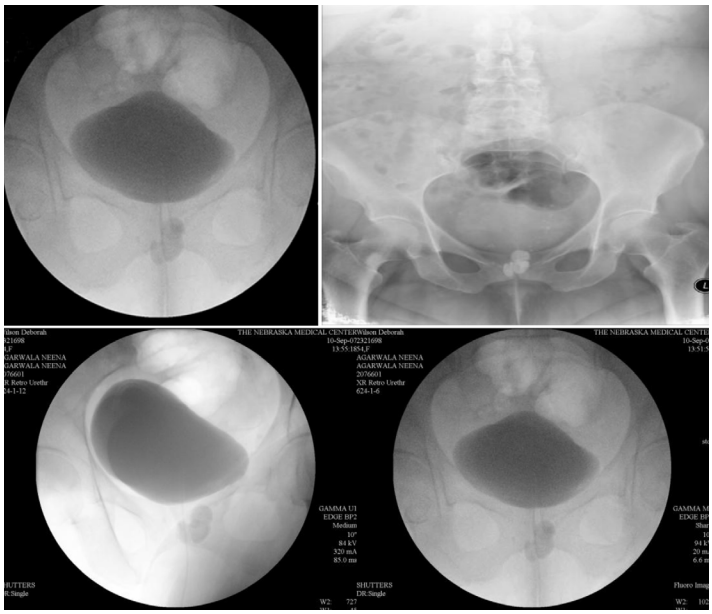

urethra. Laser removal of the stones was done prior to urethral diverticulectomy, contrary to our patient's treatment. This case report raised the possibility of decreasing urethrovaginal fistula formation through initial laser therapy [4]. The other case, involving a mulitgravida 81-year-old female, had diabetes (DM) and hypertension (HTN). Her imaging, including a plain X-ray, excretory urography, cystourethrography, and cystourethroscopy, failed to accurately diagnose due to filling defects from an obstructing calculus. Surgery indicated a stony vaginal mass that ultimately detected the diverticulum. She also had a catheter for a prolonged period of 10 days [5].

A 54-year-old, multiparous woman differed in presentation, lacking a history of UTI. She had symptoms for 1 year, analogous to our case. Urethrocytoscopy and a voiding cystourethrogram (VCUG) revealed 17 stones, which were removed and treated with a diverticulectomy. However, this patient had a Foley catheter for only 3 days and subsequently developed stress incontinence after 6 weeks for which she did not have a previous history [7].

The above cases share many of the same qualities on initial presentation. It appears there is a positive correlation between symptom duration and UTI presentation. Therefore, an early diagnosis utilizing reliable imaging may prevent UTIs from occurring, not only eliminating complications but allowing earlier surgical intervention and treatment.

Whether one prefers cheap-but-simple or expensive-but-more accurate treatment, deciding the next option may include a physical exam, sonography and/or urethroscopy, or a VCUG, especially with a double-balloon catheter (DBC) urethrogram [7]. A bimanual exam, positive for a palpable hard suburethral mass, is the most important physical diagnostic sign of diverticulum containing calculi; however, additional imaging is essential to precisely determine anatomy for surgical intervention [5]. Transrectal sonography also has accurate visualization, specifically of the diverticular wall and internal characteristics [5], and it is cheaper [3]. However, sonography is operator dependent and may not discern the diverticular neck or even differentiate between other cystic structures [2]. Cystourethrography can confirm diverticuli and associated calculi, but it is only 65% accurate when used alone because stones can obstruct the opening, preventing contrast from filling the diverticular sac [5]. Therefore, it is ideal to use VCUG with DBC to improve detection, which may otherwise be prevented by calculi or masked by the catheter because of location and positioning. DBC relies on radiologist expertise; therefore, X-ray imaging before surgery is vital in confirming physical properties, including the size of the opening and the length and number of diverticuli [7]. While CT studies accurately reveal calculi within diverticuli, most CT tests are ordered as indications for unrelated workups that frequently result in incidental diverticuli [2].

An MRI is superior to the above for preoperative imaging and reconstruction as it does not require a patent diverticular orifice and is capable of distinguishing cystic structures. To further enhance the signal-to-noise ratio and spatial resolution, endoluminal coils have recently become available [2].

Following surgical resolution and reconstruction, complications can occur in up to 5 to 46% of patients. They may either be immediate, including urethrovaginal fistulas and urethral obstruction, or delayed, including recurrent UTIs, urethral syndrome, urinary incontinence, and recurrent diverticuli [7]. In preparing treatment for our case, surgical materials were selected based on the successful repair of previous fistulas. Since the recurrence rate of fistulas is so high, the use of biological grafts, particularly xenograft, was implemented because the tissue does not need to be mobilized and is less disfiguring to the patient compared to other autologous interposition grafts. This graft—an acellular bovine collagen matrix—also decreases the risk of erosion because it is biomaterial versus synthetic, like allogeneic grafts. This may be because the use of a biomaterial is an ideal scaffold for angiogenesis and cellular growth, therefore expediting recovery of the patients and avoiding additional morbidities. Additionally, it is readily available in larger quantities, is cost-effective, and provides an excellent alternative to autologous tissue in the appropriate patient [8].

As previously discussed with laser treatment, decreasing postoperative complications may also involve increased catheter placement duration. When comparing this 54-year-old female with additional cases, it appears the length of catheter placement can alter the chance of developing problems after surgery. Because there are few cases to evaluate the length of catheter placement, future studies should be considered to confirm the impact of catheter length.

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CONCLUSION

Awareness of vague symptoms like recurrent UTIs and various urinary problems should ideally prompt clinical suspicion of urethral diverticuli. Selecting the best imaging alone or in combination with an MRI and a VCUG can guide earlier detection and prevent preoperative complications. Avoiding postoperative complications remains poorly understood and much work is yet to be done to allow sufficient intervention.

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