Case report

Urethral diverticulum with multiple calculi with presentation of urinary incontinence in a female — A case report and literature review

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1. Introduction

Urethral diverticulum (UD) with multiple calculi is a rare condition that is often difficult to diagnose. We present a 46-year-old female with initial symptoms of stress urinary incontinence (SUI) and subconscious urine leakage. Magnetic resonance imaging (MRI) revealed a large diverticulum with multiple stones in it. Transvaginal repair of the UD was performed, and her SUI subsided after diverticulectomy alone, against our expectations.

2. Case Report

A 46-year-old female presented with SUI and subconscious urine leakage for 6 months that worsened shortly prior to her admission. She had not visited other physicians for this problem although this really troubled her. The patient had to change her trousers two to three times a day, and had to use two to three pads per day. She denied dysuria, dyspareunia, other lower urinary tract symptoms, or urinary tract infection. She had three pregnancies that resulted in two natural spontaneous deliveries and one cesarean section. The result of her urinalysis was within normal limits, and the pelvic examination revealed urine leak when coughing and no continuous urine leakage. In addition, a 2 cm × 1 cm hard nodule was noted at the anterior urethra (Fig. 1). There was no discharge after urethral stripping. A cystourethroscopy did not reveal any specific finding. Her MRI scan revealed a 2-cm UD at the distal urethra 3 cm away from the bladder neck with four well-defined stones, and the wall of the diverticulum was smooth (Fig. 2). We suggested transvaginal urethral diverticulectomy with concomitant autologous pubovaginal fascial sling, but the patient decided not to undergo anti-incontinence surgery because of infection risk and postoperative pain. The patient was placed in a lithotomy position. The stones were exposed with a longitudinal incision (Fig. 3) to the ostium communicating with the diverticulum to the urethral lumen. An en bloc excision was performed, and all stones were removed (Fig. 4). Multilayered, watertight closure of the urethra and vagina wall was performed with absorbable sutures. At 2 weeks after the operation, the wound was well healed and her incontinence subsided after diverticulectomy alone. Her condition did not change at 6 months after the operation, against our expectations.

3. Discussion

UD was first reported 2 centuries ago by Hey,1 but until the invention of positive-pressure urethrography in 1952 by Davis and Cian,2 only a few cases were discovered. Up to 20% of patients may be asymptomatic; therefore, the true incidence is unclear because...
of missed diagnoses, although the accepted prevalence is about 0.5–6%.

UD patients present with diverse symptoms, but the classical symptoms of “3Ds” are dysuria, dyspareunia, and postvoiding dribbling. Other symptoms associated with UD are frequency, urgency, hematuria, SUI, and persistent pyuria. The most important findings on physical examination are palpable vaginal mass and discharge after stripping. However, in a series with 46 female patients with urethral diverticula, the mean time from occurrence of symptoms to diagnosis is 5.2 years, despite the fact that 52% of those patients had palpable vaginal masses—as in the case of our patient, who did not notice the vaginal mass but sought medical advice because of symptoms of SUI.

Double balloon positive pressure urethrography is a good diagnostic tool but is not widely available. Compared to voiding cystourethrography and ultrasonography, high-resolution MRI has higher sensitivity and specificity, especially when differential diagnoses of urethral masses other than UD are present.

UD presenting with urolithiasis is uncommon, with an incidence of 1–10%. The etiology may be deposition, stasis of infected urine, or stone migration. To our knowledge, studies seldom compare the diagnosis and management between UD with or without stones. Almost all UD with stone present with palpable hard vaginal masses, and patients tend to receive surgical excision rather than conservative treatment.

The management of UD depends on whether it is symptomatic. Low-dose antibiotics and urethral stripping after voiding may be used in asymptomatic patients or patients with smaller diverticula.

Some case reports revealed that malignancies such as squamous cell carcinoma, adenocarcinoma, and clear cell carcinoma may originate from the diverticulum, but the risk is very low, and only about 100 cases have been reported so far. This should be made known to the patient, and regular follow-up should be advised.

For symptomatic patients, surgical repair is indicated. Complete excisional diverticulectomy with multilayer watertight closure showed the highest efficacy in most patients with small defects. In patients with larger defects, poor tissue quality, or suboptimal blood supply, Martius flap is a reliable procedure with a low complication rate. Up to 30% of patients have SUI prior to diverticulectomy, and about 15–49% had de novo SUI after surgery.

Fig. 1. A 2 cm × 1 cm hard nodule over the anterior urethra.

Fig. 2. Four smooth stones inside the urethral diverticulum.

Fig. 3. En bloc excision was performed and the urethral wall repaired.

Fig. 4. All the stones were removed.
surgery. For patients with UD and preoperative SUI, anti-incontinence surgery such as pubovaginal fascial sling resulted in a significant decrease in postoperative SUI. The use of slings is safe and does not increase infection risk, and it is considered the “gold standard” treatment modality. The synthetic mid-urethral sling used for this condition is not well studied, and most authors would avoid using it owing to the potentially increased infection risk. Our patient had a large UD with stones with concomitant SUI, and she opted not to undergo anti-incontinence surgery. However, her SUI was cured after diverticulectomy, which was against our expectations. In another series, 89 patients with UD received diverticulectomy with or without Martius flap but not anti-incontinence surgery; 32 patients had preoperative SUI, and 20 of them were cured after diverticulectomy without sling insertion. De novo SUI was noted in 13 patients, and a total of 25 patients had postoperative SUI. Twelve out of 25 patients had mild symptoms and did not require further management. Another study of 50 patients with UD reported an incidence of de novo SUI rate as 49%, but most patients had mild symptoms and only five patients required subsequent anti-incontinence surgery. The mechanism of how diverticulectomy cures SUI is unclear. One hypothesis is that a large diverticulum may compromise urethral support and cause SUI. Another hypothesis is that the pre-operative SUI reported in certain patients was not true SUI, but was actually paradoxical stress incontinence, that is, the loss of retained urine in the diverticulum during coughing. In conclusion, in patients with UD and concomitant SUI, diverticulectomy with anti-incontinence surgery is accepted and performed by most clinicians; however, staged operation is another alternative that may prevent unnecessary treatment.

Conflicts of interest

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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