

# Urethral diverticular with broadly squamous metaplasia in a patient with urethral diverticular calculi

## A case report

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### Abstract

**Rationale:** There are already several reports concerning the occurrence of urethral diverticulum (UD) in female patients, but only rarely has an article describing UD combined with UD calculi or squamous carcinoma been published. Moreover, a case with squamous carcinoma and UD calculi at the same time has never been reported, making this the first case report about this condition.

**Patient concerns:** A 43-year-old woman presented to the gynaecology department with a complaint of a hard mass beneath the anterior vaginal wall.

**Diagnoses:** Transvaginal ultrasound (TVU) revealed a UD.

**Interventions:** We performed a standard urethral diverticular excision. Intraoperatively, we identified and removed a stone from the diverticulum. The intraoperative finding of a stone challenged the diagnosis of UD, with subsequent histological examination of biopsy tissue from the mass demonstrating broadly squamous metaplasia.

**Outcomes:** The broadly squamous metaplasia predominantly originated from the stone, and the stone was entirely removed. No complications occurred during the whole follow-up period. Moreover, after the 12-month follow-up, there was no diverticular recurrence or carcinoma metastasis.

**Lessons:** UD calculi may be considered a risk factor for female urethra squamous metaplasia, which with the potential of squamous carcinoma, so patients will be advised to treat this condition immediately.

**Abbreviations:** MRI = magnetic resonance imaging, TVU = transvaginal ultrasound, UD = urethral diverticulum.

**Keywords:** broadly squamous metaplasia, calculi, urethral diverticular

## 1. Introduction

Squamous carcinoma has a reported prevalence of 5% in female patients with urethral diverticulum (UD),<sup>[1]</sup> with UD calculi also rarely reported.<sup>[2]</sup> The primary approach to treating UD is surgical resection. Even though several surgical techniques have

been described with transvaginal diverticulectomy<sup>[3,4]</sup>; however, some common complications, such as the coexisting stone and accompanying carcinoma, may occur without drawing enough attention, making it challenging to study them prospectively. Herein, we report a case of UD with broadly squamous

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YS, CT, and Y-jB contributed equally to this work and should share the co-first author.

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**Consent section of patient:** Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

**Availability of data and materials:** Those data supporting our findings can be found in the His system of West China Hospital.

The authors report no conflicts of interest.

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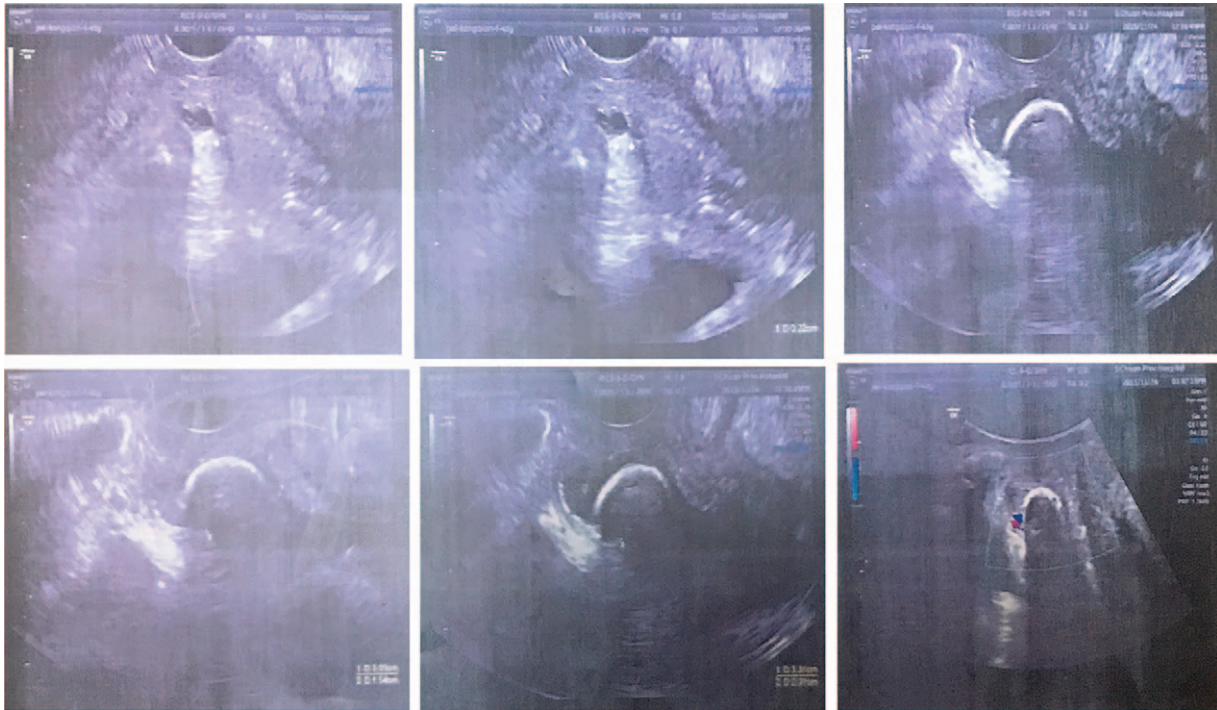
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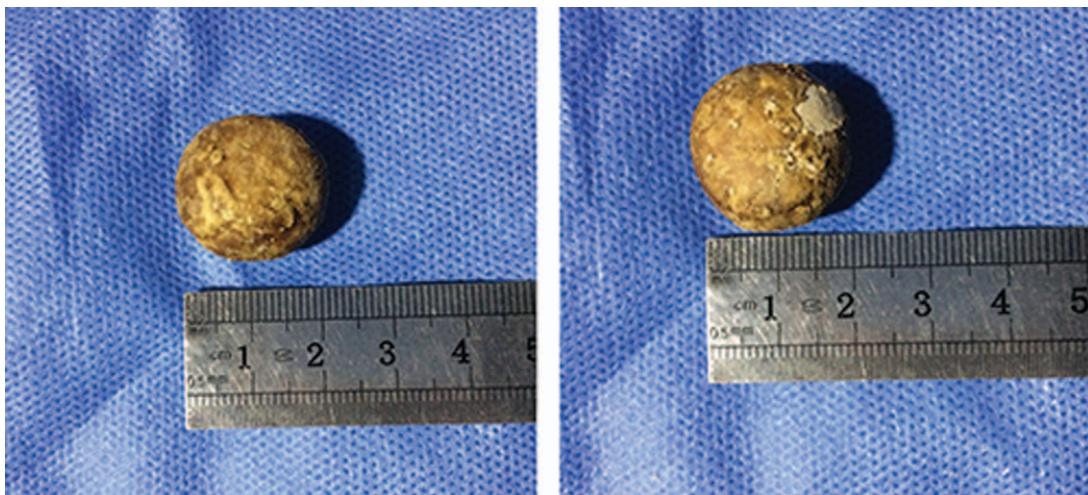
**Figure 1.** The result of transvaginal ultrasound examination. Transvaginal ultrasound revealed a urethral diverticular but did not find the stone.

metaplasia (which with the potential of squamous carcinoma) in a patient with UD calculi.

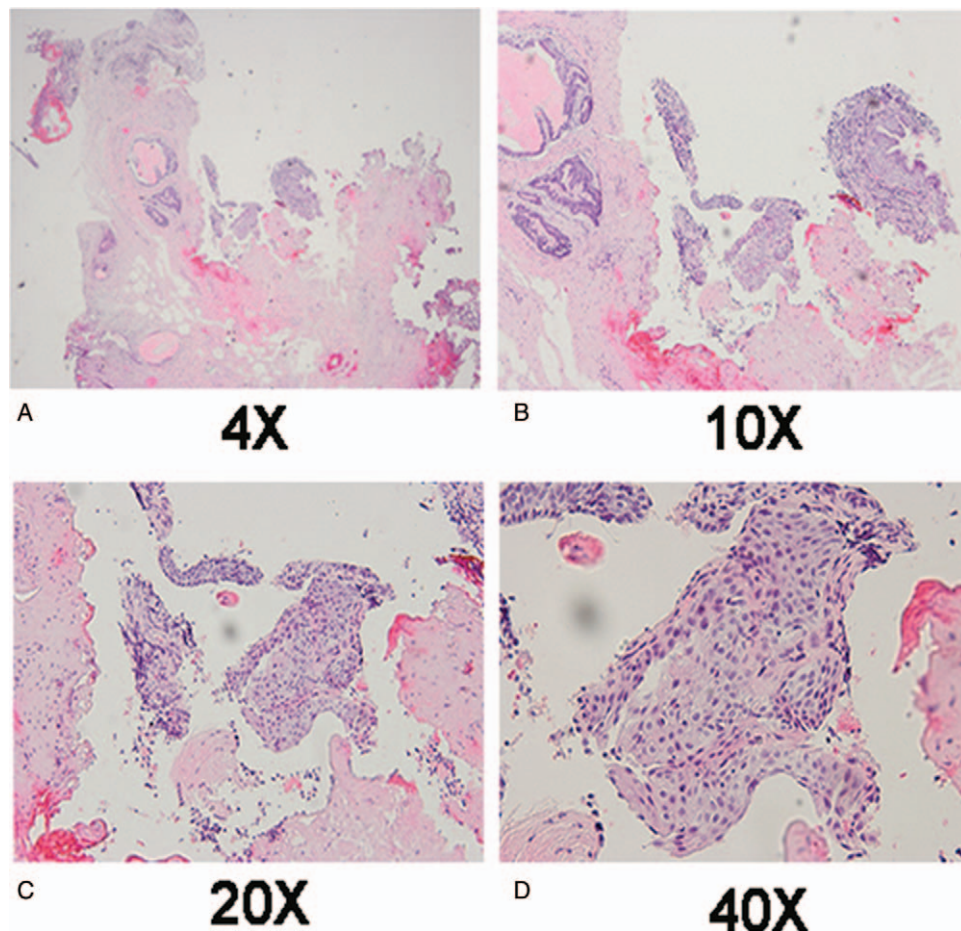
## 2. Case report

A 43-year-old woman presented to the urogynaecology department with a hard mass beneath the anterior vaginal wall. Vaginal examination revealed a hard mass located 2.5 cm from the orificium urethrae externum and beneath the anterior vaginal wall. Transvaginal ultrasound (TVU) revealed a cystic mass with calcification at the front and right rear walls of the urethra (Fig. 1). Based on the diagnosis of UD, the patient was referred to

our department. The patient had first become aware of the mass 2 months earlier, with accompanying symptoms of dyspareunia and without any urination symptoms or bleeding upon compression of the mass. On admission, we performed a standard UD excision.<sup>[1]</sup> And then, we identified in the process of operation and removed a stone from the diverticulum. Macroscopic examination of the resected specimen revealed that the UD was located at the left side of the urethra and ranged from 4 o'clock to 7 o'clock with a stone of 2.0 cm in diameter (Fig. 2). Histopathological examination of the UD wall demonstrated broad squamous metaplasia and high-level intraepithelial neoplasia within the mucous epithelium, with evidence of



**Figure 2.** Photograph of the urethral diverticular calculi. The diameter of this stone is approximately 2 cm.



**Figure 3.** Pathological section of the urethral diverticular wall. Histopathological examination of the urethral diverticular wall revealed a broadly squamous metaplasia and a high-level intraepithelial neoplasia in the mucous epithelium and that a preinvasive carcinoma has been formed. (A) Four times enlarged; (B) 10 times enlarged; (C) 20 times enlarged; (D) 40 times enlarged.

pre-invasive carcinoma, still in the stage of TisN0M0 (Fig. 3), and the resected margins are negative. Because the patient had an uneventful postoperative course and reported no particular discomfort, the patient was discharged from our department on postoperative day 7. The patient was recovering well without the diverticular recurrence or carcinoma metastasis at the 12-month follow-up. Moreover, there were no complications when this patient was in the hospital and at the follow-up. In the whole follow-up period, both the carcinoembryonic antigen and alpha fetoprotein are normal.

### 3. Discussion

UD carcinoma is a rare tumor affecting female patients that was first reported in 1951.<sup>[5]</sup> Less than 60 cases of UD carcinoma, including adenocarcinoma and squamous cell carcinoma, have been reported.<sup>[6]</sup> The pathogenic mechanisms underlying the development of UD carcinoma are poorly understood because of a lack of case reports.<sup>[7]</sup> In fact, with regards to the pathology results presented, a 6% rate of adenocarcinoma was quoted by Thomas et al<sup>[8]</sup> in a larger study population (90 patients), and those patients with carcinoma remained tumor-free for 2 years.

In the present case, UD carcinoma was accompanied by a stone. According to previous reports, UD stones may result from

chronic stasis and urinary infection within UD or proximal to a urethral obstruction.<sup>[9,10]</sup> Although the present case of UD carcinoma with associated calculi is an extremely rare condition, further studies are required to increase our clinical understanding of UD and improve patient outcomes.

The most common presenting symptom of UD is urethral bleeding, which occurs in 51% of patients, with other nonspecific lower urinary symptoms frequently reported, such as increased frequency, urgency and localized pain.<sup>[7]</sup> Urinary retention is observed in 4% of cases of female patients.<sup>[11]</sup> However, the most prominent symptom was dyspareunia without bleeding in the present case. Magnetic resonance imaging (MRI) plays an important role in the imaging of UD<sup>[9]</sup>; however, stones are not readily identifiable on MRI. A recent study demonstrated the superior efficacy of TVU compared with MRI<sup>[12]</sup>; however, TVU did not identify the presence of a stone in the present case. In addition, physical examination is commonly performed in patients suspected to have UD stones, with the identification of a crackling mass on pelvic examination considered highly suggestive.<sup>[2]</sup> In the present case, vaginal examination revealed a hard mass located 2.5 cm from the orificium urethrae externum and beneath the anterior vaginal wall. Excision of UD only was performed in the present case because of the increased risks and higher recurrence rates associated with complete vaginal

excision.<sup>[4,12]</sup> Long-term follow-up for diverticular recurrence and carcinoma metastasis by physical examination and imaging is required in the present case despite the success of conservative surgery with few complications because of the intraoperative identification of a UD calculus and subsequent demonstration of a UD carcinoma by pathological examination. Subsequently, UD calculi may be considered a risk factor for squamous metaplasia, which with high potential of squamous carcinoma in the female urethra, and UD calculi may be considered an indication that surgery is necessary. However, further study is necessary.

### Author contributions

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