

## Case Report

# Urinary retention in adult hypospadias patient resolved by unroofing of a mildly enlarged prostatic utricle

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### Abbreviations & Acronyms

CT = computed tomography  
MRI = magnetic resonance imaging  
TRUS = transrectal ultrasonography  
VCUG = voiding cystourethrogram

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**Introduction:** A prostatic utricle is a congenital saccular indentation in the prostatic urethra and frequently enlarged in hypospadias patients. We present a case of urinary retention associated with a mildly enlarged utricle.

**Case presentation:** A 20-year-old male, who underwent multiple repair procedures for hypospadias during childhood, was referred to us for dysuria. Retrograde urethrogram, voiding cystourethrogram, and cystoscopy results revealed only a mildly enlarged prostatic utricle, with no apparent lower urinary tract obstruction or urethral valves. A meatotomy was performed under suspicion of meatal stenosis, though urinary retention occurred following that procedure. Transrectal ultrasonography revealed flapping of the prostatic urethra floor over the utricle. Transurethral unroofing of the utricle relieved the dysuria.

**Conclusion:** A mildly enlarged prostatic utricle can cause dysuria. To the best of our knowledge, no case similar to the present has been previously reported.

**Key words:** dysuria, hypospadias, prostatic utricle, transurethral surgery, transrectal ultrasonography.

## Keynote message

Mildly enlarged prostatic utricles can cause dysuria. TRUS is a useful imaging tool for visualizing dynamic movements of the urethral floor over the utricle.

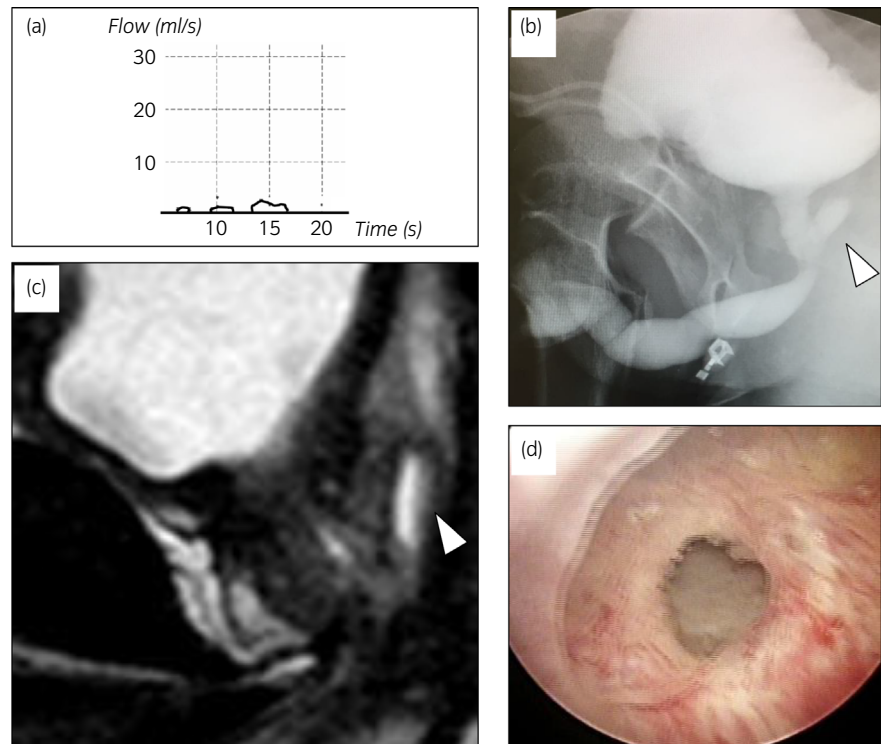
## Introduction

Enlarged prostatic utricle is a congenital saccular indentation in the prostatic urethra, formerly known as “vagina masculina” or “uterus masculinus.”<sup>1</sup> The utricle is considered to be a remnant of a Müllerian duct or urogenital sinus, and reported to be visualized by urethrogram in 31.5% of hypospadias cases.<sup>2</sup> Enlargement of prostatic utricle is classified into four grades according to extension of the lesion,<sup>3</sup> with one that shows significant enlargement classified as Grade 2 or 3, and known to sometimes cause dysuria, hematuria, or infection.<sup>4–6</sup> We present here a case of dysuria associated with prostatic utricle with milder (Grade 1) enlargement, in an adult patient who underwent multiple hypospadias repairs during childhood.

## Case presentation

A 20-year-old male was referred to us for dysuria. He had previously undergone an initial two-stage repair for scrotal hypospadias at the age of 4 years at our hospital, after which reduction urethroplasty for diverticular enlargement of the urethra was performed at 12 years as well as several internal urethrotomy procedures at another hospital. At presentation, his urethral meatus existed at the tip of the glans, and he continued self-dilation of the urethral meatus by catheterization, though improvement in dysuria was not noted. Uroflowmetry test results obtained at the initial visit showed a very poor urinary stream (Fig. 1a).

Retrograde urethrogram and VCUG revealed a mildly enlarged prostatic utricle, Grade 1, with no apparent urethral stricture (Fig. 1b). MRI also showed the prostatic utricle (Fig. 1c),



**Fig. 1** (a) Preoperative uroflowmetry results indicating severe dysuria. (b) Preoperative VCUg showing Grade 1 enlargement of prostatic utricle (arrow) and stenosis of repaired urethral orifice. (c) High-intensity signal in T2-weighted MRI demonstrating intracystic contents of prostatic utricle (arrow). (d) Preoperative cystoscopy image demonstrating orifice of prostatic utricle.

but no spinal malformation. Urodynamic study findings indicated that bladder capacity, filling and voiding phase pressure, and bladder sensation were normal. Cystoscopy showed no posterior urethral valves, though the orifice of the prostatic utricle was seen located just proximal to the sphincter (Fig. 1d).

A meatotomy was performed to release the suspected meatal stenosis. However, dysuria worsened, and recurrent episodes of urinary retention were noted following the procedure. Management by intermittent catheterization was not possible, because the catheter tip easily migrated into the utricle and a suprapubic cystostomy tube was indwelled. However, he urinated quite easily when a thin urodynamic catheter was inserted into the urethra, which made us suspect the presence of a flap-like mechanism.

We decided to unroof the prostatic utricle to facilitate catheterization and micturition.

At the beginning of surgery, TRUS was performed to dynamically visualize the utricle (Fig. 2a). Micturition maneuver was simulated by manual compression of the fully distended bladder (Fig. 2b). The utricle was unroofed by resecting the posterior urethral wall. The resection began from the orifice of the utricle and continued up to the bladder neck (Fig. 3a,b).

Postoperatively, urine stream was dramatically improved beyond our expectation. Additionally, VCUg revealed smooth urination (Fig. 3c), with a maximum flow rate in a uroflowmetry test of 30 mL/s and no residual urine (Fig. 3d).

## Discussion

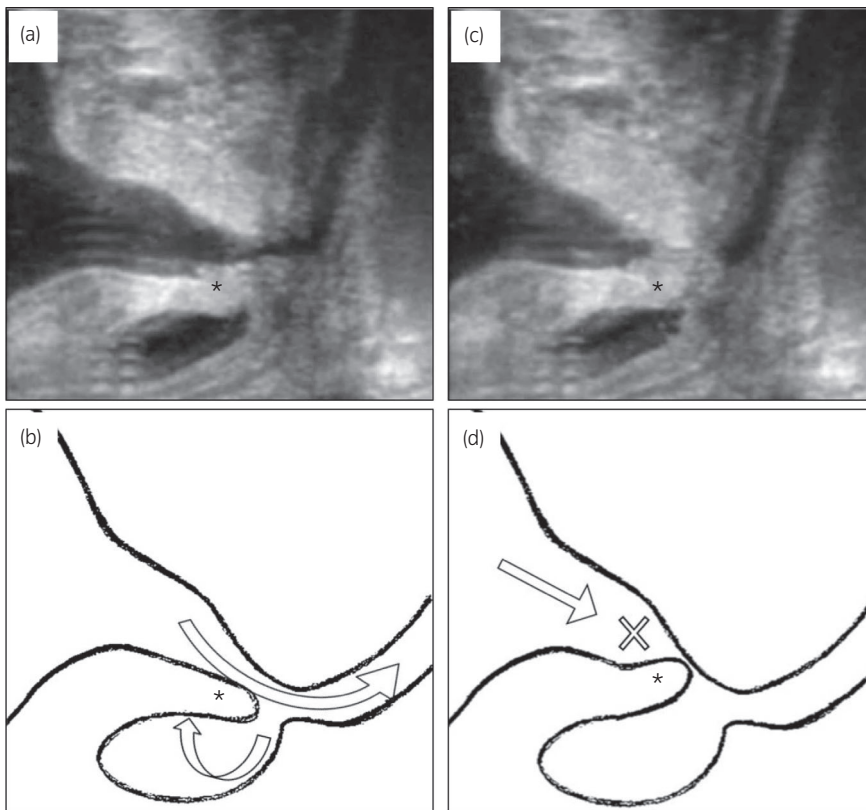
A Müllerian duct remnants in the prostate can be either an isolated cystic lesion, known as a Müllerian duct cyst, or

saccular indentation communicating with the urethra just distal to the verumontanum, termed prostatic utricle.<sup>1,7</sup>

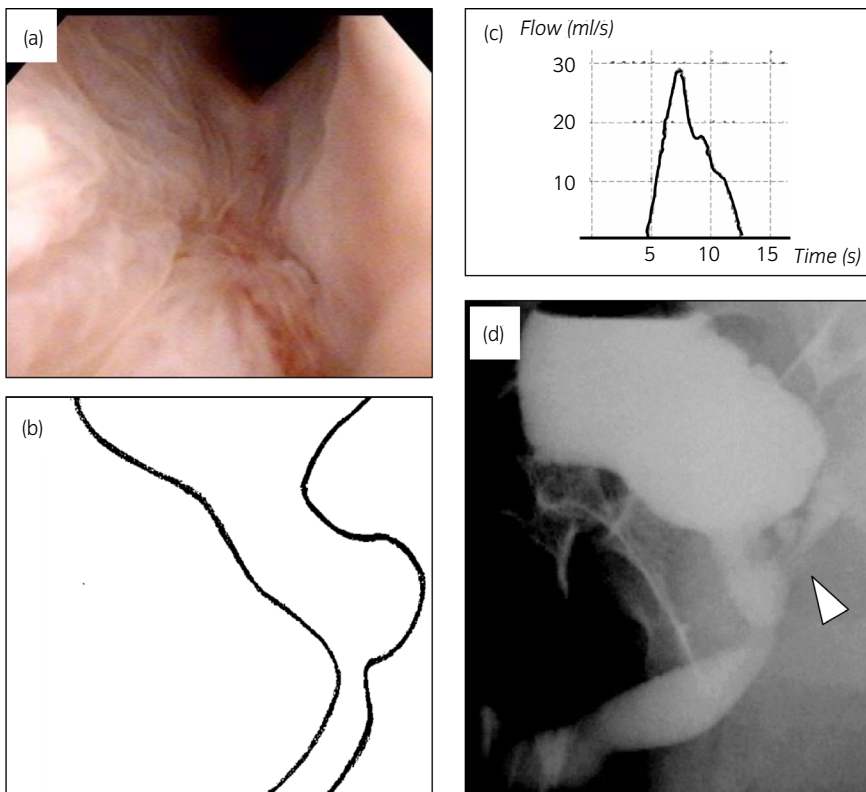
Enlargement of prostatic utricle is classified according to location, from Grade 0 to 3 (Figs S1 and S2).<sup>3</sup> Those with larger prostatic utricle (Grade 2 and 3) present various clinical findings,<sup>4</sup> but smaller prostatic utricle (Grade 0 or 1) is often asymptomatic and needs no treatment.<sup>2,3,5,6</sup>

The present case is an exceptionally rare presentation, in which a Grade 1 mildly enlarged prostatic utricle was associated with urinary retention. A search of literature did not find a similar case. Diaz *et al.* reported a case of Müllerian duct remnants, in which transurethral resection of the prostatic cyst wall along with resection of the prostate improved the quality of life by preventing urinary retention. The authors concluded that the cyst had obstructed the bladder outlet with a ball-valve mechanism.<sup>8</sup> In the present case, urinary retention did not seem to be caused by a ball-valve mechanism. TRUS performed during manual abdominal compression revealed a flapping motion of the floor of the prostatic urethra over the utricle (Fig. 2c,d). The manual compression is not identical to normal voiding reflex, but the urine stream was flowed into the utricle through the orifice. The mechanism of the flapping motion was related to fragility of the prostatic urethra floor. We assumed the fragile floor was elevated by the urine stream flows into the utricle during urination, and the pushed-up floor inhibited urination. For any reason, unroofing of the utricle by transurethral resection resulted in dramatically improved urination.

Various surgical procedures to treat a symptomatic enlarged prostatic utricle have been described. Laparoscopic or robotic excision affords minimally invasive access to the retrovesical space.<sup>9,10</sup> However, such resection of the



**Fig. 2** (a) Transrectal ultrasound showing prostatic utricle (\*) at beginning of manual abdominal compression. (b) Schema for mechanism of backward urine flow into prostatic utricle during manual abdominal compression. (c) Transrectal ultrasound demonstrating flapping motion of posterior wall of prostatic urethra (\*) during manual abdominal compression. (d) Schema for mechanism of elevation of prostatic urethra (\*) posterior wall by backflow, causing obstruction of lower urinary tract.



**Fig. 3** (a) Prostatic urethra after 3 months from unroofing of prostatic utricle. (b) Schema for sagittal section of prostatic urethra after unroofing of prostatic utricle. (c) Postoperative uroflowmetry indicating normal urination. (d) Postoperative VCUG demonstrating smooth urination after unroofing of prostatic utricle (arrow).

utricle should only be partial and avoid destruction of the seminal tract, and its indication is limited to larger utricles.<sup>8</sup>

Transurethral endoscopic unroofing of the utricle, consisting of orifice dilation, catheterization and aspiration, and resection, is a less-invasive procedure, though associated

with a relatively high risk of recurrence in cases with a larger utricle.<sup>5</sup> Although we anticipated, though were not extremely confident, that relief of obstruction could be obtained in this case, the procedure was primarily chosen for easier catheterization. Presently, the patient is able to undergo catheterization at a local clinic or perform it by himself without difficulty. One drawback of that procedure is that it may cause retrograde ejaculation. Our patient did not document ejaculation experience prior to the operation and gave consent to unroofing the utricle because of the minimal invasiveness.

Pelvic ultrasonography, TRUS, and perineal ultrasonography can each show the prostate and urethra.<sup>3,11</sup> In the present case, TRUS was able to effectively delineate the dynamic motion of the prostatic floor and roof of the utricle, and was considered to be the most useful tool for delineating the dynamics of the urethral floor.

## Conclusion

A slightly enlarged prostatic utricle could be a cause of dysuria, which was resolved by a transurethral unroofing procedure. TRUS was useful for determining diagnosis.

## Conflict of interest

The authors declare no conflict of interest.

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## Supporting information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

**Figure S1.** Grade 0: Utricle opening located on urethra, but not extending over verumontanum. Grade 1: Utricle larger than Grade 0, but does not reach to bladder neck. Grade 2: Utricle shows greater enlargement and dome extending over bladder neck. In Grade 0, 1, and 2 cases, the utricle opens in the prostatic urethra proximal to the sphincter. Grade 3: Opening situated in bulbous urethra just distal to sphincter.

**Figure S2.** Preoperative urodynamics data showing normal filling and voiding phase.