



## CASE REPORT

# Congenital duplication of the urethra with urethral diverticulum: a case report [v1; ref status: indexed, <http://f1000r.es/36h>]

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## Abstract

Duplication of the urethra is a rare congenital anomaly. Urethral duplication with the presence of diverticulum is a rare combination and to the best of our knowledge has not been previously reported. We report a case of a 16 month old male child with duplication of the urethra and diverticulum arising from the ventral urethra. We also cover the intricacies and challenges in the management of such a case.

The opening of the narrowed accessory dorsal urethra at the verumontanum was cauterized and gradually the dorsal urethra became atrophied. The ventral urethral diverticulum was excised. This case is unique due to:

- The unusual presentation of swelling over the dorsum of the penis, together with duplication of the urethra with diverticulum.
- The use of cauterization as a treatment modality. Cauterization of the ventral urethra with a Bugbee electrode and diverticulectomy was performed. A glidewire helped in identifying the small opening of the dorsal urethra at the level of the verumontanum.

The case also highlights the importance of endoscopic management of this clinical entity.

## Open Peer Review

### Invited Referee Responses

	1	2	3
<b>version 1</b> published 01 May 2014	report	report	report

- 1 **Ali Tourchi**, The Johns Hopkins Medical Institutions USA
- 2 **Sanjay Kulkarni**, Centre for Reconstructive Urethral Surgery India, **Pankaj Joshi**, Centre for Reconstructive Urology India
- 3 **Tamsin Greenwell**, University College London Hospital UK

### Latest Comments

No Comments Yet

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## Introduction

Duplication of the urethra is a rare congenital anomaly. Most cases involve incomplete duplication of the urethra. This anomaly is more common in males<sup>1</sup>. The etiology of urethral duplication is unclear, no hypothesis explains the basis for all cases<sup>2,3</sup>. Urethral duplication with the presence of a diverticulum is a rare combination and to the best of our knowledge has not been previously reported.

In this report we describe a rare case of urethral duplication presenting as a urethral diverticulum in the ventral urethral passage, whilst the dorsal opening was abnormal. We also allude to the intricacies and challenges in the management of such a case.

## Case report

A 16 month old male child from India with duplication of the urethra and a diverticulum arising from ventral urethra was presented at our clinic in 2013. He presented with swelling of the penis, which increased in size with urination. General examination revealed an otherwise healthy child. Local examination revealed an approximately 4x2cm sized soft tissue swelling on the ventral aspect of the distal part of penis, cystic in nature (Figure 1A). The meatus at the tip of the glans (dorsal urethral opening) of the penis was tiny and admitted a no. 22 G Intracath tip. On compression, drops of urine egressed from the meatal opening. Another meatal opening (ventral urethral opening) was seen 10mm proximal and ventral to first opening, which was wide and admitted a 10fr (3.33mm) infant feeding tube. A micturating cystourethrogram (MCUG) showed complete duplication of the urethra with a diverticulum arising from ventral urethra near its terminal part with a small para ureteral bladder diverticulum (Figure 1B).

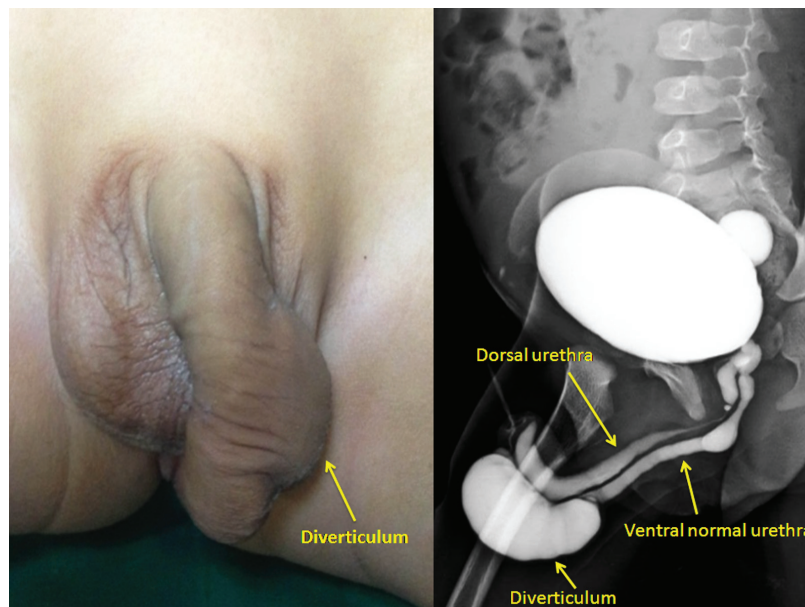
Cystoscopy was done with a 9.5Fr cystoscope (KarlStorz, Germany). A Bugbee electrode, 3fr (KarlStorz, Germany) that could pass through a 3fr working channel of the cystoscope was used. Cystoscopy revealed a normal ventral urethra with a diverticulum. The dorsal urethra was abnormal and was narrowed in the proximal part. Its opening into the normal ventral urethra was localized with difficulty after passing a 0.025"/0.64mm glidewire (Terumo Corporation, Tokyo-Japan) through it (Figure 2A). The opening was just proximal to the verumontanum at 10 o'clock. The dorsal urethra was cauterized at its opening into the ventral urethra using a Bugbee electrode and open diverticulectomy of the ventral urethral diverticulum was performed.

The patient was doing well at 6 months follow up with a good urinary stream (Figure 2B).

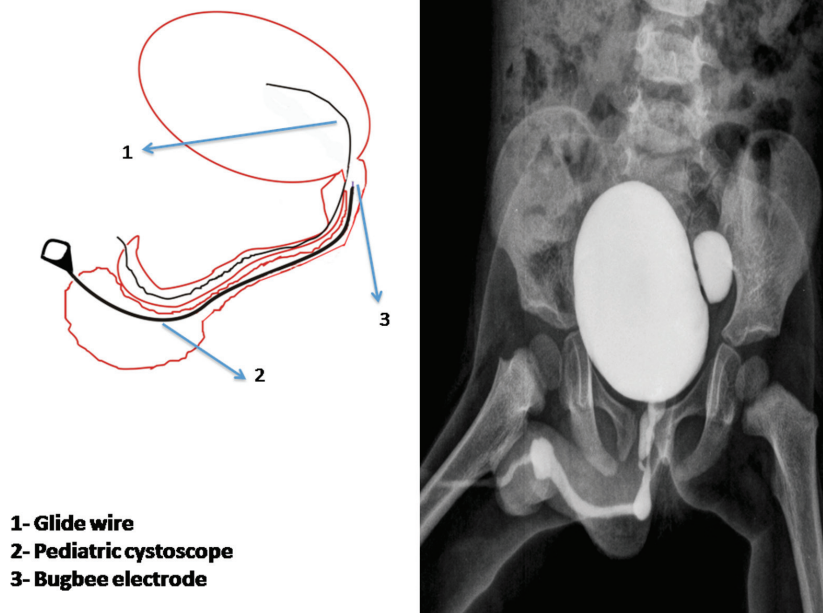
## Discussion

Duplication of the urethra can occur with complete duplication of the penis or urinary bladder in the most extreme cases<sup>4</sup>. Urethral duplication may be sagittal or collateral. In our case it was sagittal. Sagittal duplication takes the form of two channels running one above the other in the sagittal plane, whereas in the collateral form, the duplicate urethras run side by side. Most urethral duplications occur in the sagittal plane within a single penis and most are incomplete. Usually in such cases the ventral urethra is the dominant one<sup>5</sup>. The most common sagittal variety is an orthotopic principal urethral channel and an epispadiac accessory urethra lying dorsal to it.

There are several different classifications describing urethral duplications. The classification by Effman *et al.* is the most widely used<sup>6</sup>.



**Figure 1.** 1A) Clinical photograph showing diverticulum at the distal part of the penis. 1B) Micturating cystourethrogram showing complete duplication of the urethra in the sagittal plane. Large diverticulum arising from distal part of ventral urethra with small bladder diverticulum.



**Figure 2. 2A)** Pictorial diagram showing the glide wire passed from the dorsal urethra to identify its opening into the normal ventral urethra. The cystoscope was passed from the ventral urethra up to the opening of dorsal urethra with a Bugbee electrode for fulguration of this opening. **2B)** Post-operative micturating cystourethrogram showing normal urethra, no urethral diverticulum and complete obliteration of the dorsal urethra is noted.

According to this classification; the present case was type II A-2 (complete urethral duplication with the second urethra arising from first one and coursing independently into separate meatus).

Clinical presentation varies from type to type. Double urinary stream is one of the presentations of urethra duplication and may be bothersome when the ventral meatus is too proximal over the penis. Presentation may also include repeated urinary tract infections, incontinence or it may be asymptomatic and the only concern being a double meatus<sup>7</sup>. This happens particularly when both meatus are very nearby.

Clinical examination and retrograde urethrogram (RGU) with MCUG should be sufficient for diagnosis in most cases. However, sonourethrograms and magnetic resonance imaging (MRI) are now also being used as adjunct procedures. Both will give excellent soft tissue details such as plaque or calcification which are associated with chordee in such cases<sup>8</sup>.

Detailed knowledge of urethral duplication is important when planning for any surgical procedure for its correction. Many patients are asymptomatic and do not require any surgery. Indications for surgery are bothersome symptoms and cosmetic or functional deformity. Surgical reconstruction varies from case to case. It may range from simple meatoplasty to complex staged urethroplasty, depending on the severity of case. Most procedures involve excision of the accessory urethra with reconstruction of the dominant urethra<sup>9</sup>. A favorable outcome is achieved in most of cases after reconstructive surgery. Dilatation of the orthotopic urethra is a more controversial

option<sup>10</sup>. Holst *et al.* have described fulguration of an atypical urethra as another treatment option<sup>11</sup>.

Our treatment technique in this case was unique (minimally invasive) and successful. The opening of narrowed accessory urethra at the verumontanum was cauterized and the dorsal narrow accessory urethra gradually atrophied and had disappeared at a 6 month follow up MCUG. The ventral urethral diverticulum was excised at same time. This approach was chosen, given the concerns of infertility and incontinence associated with excision of such a long abnormal urethral tract<sup>5</sup>.

In summary, the uniqueness of our case lies in the following facts:

- Unusual presentation of swelling over the dorsum of the penis, duplication of urethra with diverticulum.
- Cauterization was used as a treatment. Cauterization of the ventral urethra with a Bugbee electrode and diverticulectomy was offered as a treatment modality. The glide wire helped in identifying the small opening.

The case also highlights the importance of endoscopic management of this clinical entity.

### Consent

Before surgical procedure written informed consent obtained from patient's parents. Written informed consent for publication of clinical details and clinical images was also obtained.

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## Author contributions

DHS and APG prepared the initial case report and literature review. RBS and MRD were involved in critical interpretation and revision of the manuscript. All authors have agreed to the final content of the case report.

## Competing interests

No competing interests were disclosed.

## Grant information

The author(s) declared that no grants were involved in supporting this work.

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## References

- Pippi Salle JL, Sibai H, Jacobson AI, *et al.*: **Bladder exstrophy associated with complete urethral duplication: a rare malformation with excellent prognosis.** *J Urol.* 2001; **165**(6 Pt 2): 2434–37.  
[PubMed Abstract](#) | [Publisher Full Text](#)
- Das S, Brosman SA: **Duplication of the male urethra.** *J Urol.* 1977; **117**(4): 452–54.  
[PubMed Abstract](#)
- Casselmann J, Williams DI: **Duplication of the urethra.** *Acta Urol Belg.* 1996; **34**(4): 535–41.  
[PubMed Abstract](#)
- Urakami S, Igawa S, Shiina H, *et al.*: **Congenital collateral urethral duplication in the frontal plane.** *J Urol.* 1999; **162**(6): 2097–98.  
[PubMed Abstract](#) | [Publisher Full Text](#)
- Salle JL, Sibai H, Rosenstein D, *et al.*: **Urethral duplication in male: review of 16 cases.** *J Urol.* 2000; **163**(6): 1936–40.  
[PubMed Abstract](#) | [Publisher Full Text](#)
- Effmann EL, Lebowitz RL, Colodny AH: **Duplication of the urethra.** *Radiology.* 1976; **119**(1): 179–85.  
[PubMed Abstract](#) | [Publisher Full Text](#)
- Erdil H, Mavi A, Erdil S, *et al.*: **Urethral duplication.** *Acta Med Okayama.* 2003; **57**(2): 91–3.  
[PubMed Abstract](#)
- Bhadury S, Parashari UC, Singh R, *et al.*: **MRI in congenital duplication of urethra.** *Indian J Radiol Imaging.* 2009; **19**(3): 232–34.  
[PubMed Abstract](#) | [Publisher Full Text](#) | [Free Full Text](#)
- Podesta ML, Medel R, Castera R, *et al.*: **Urethral duplication in children: surgical treatment and results.** *J Urol.* 1998; **160**(5): 1830–33.  
[PubMed Abstract](#) | [Publisher Full Text](#)
- Ortolano V, Nasrallah PF: **Urethral duplication.** *J Urol.* 1986; **136**(4): 909–12.  
[PubMed Abstract](#)
- Holst S, Peterson NE: **Fulguration-ablation of atypical accessory urethra.** *J Urol.* 1988; **140**(2): 347–48.  
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# Open Peer Review

Current Referee Status:



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## Referee Responses for Version 1



**Tamsin Greenwell**

Institute of Urology, University College London Hospital, London, UK

**Approved: 20 August 2014**

**Referee Report:** 20 August 2014

**doi:**[10.5256/f1000research.4121.r5660](https://doi.org/10.5256/f1000research.4121.r5660)

This is a nicely written case report of a novel treatment of an unusual problems and deserves indexing.

**I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.**

**Competing Interests:** No competing interests were disclosed.



**Sanjay Kulkarni<sup>1</sup>, Pankaj Joshi<sup>2</sup>**

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**Approved: 08 August 2014**

**Referee Report:** 08 August 2014

**doi:**[10.5256/f1000research.4121.r5659](https://doi.org/10.5256/f1000research.4121.r5659)

This is a good case report.

It highlights the fact that urethral duplication has varied presentations.

Ventral diverticulum actually helped in diagnosing the patient early as it was a symptomatic cosmetic presentation.

The authors are highly skilled in Endourology and skillfully performed the cauterization of the dorsal urethra.

This suggests that minimally invasive endourological technique can be used for treatment of duplication.

The follow-up MCU shows excellent result.

**We have read this submission. We believe that we have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.**

**Competing Interests:** No competing interests were disclosed.



**Ali Tourchi**

Division of Pediatric Urology, The Johns Hopkins Medical Institutions, Baltimore, MD, USA

**Not Approved: 01 August 2014**

**Referee Report:** 01 August 2014

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The case reported in this paper is an extremely rare presentation of urethral duplication which led me to read it three times. Urethral duplication is a rare congenital finding in pediatric urology. Most patients with this anomaly are asymptomatic. When they are symptomatic, they present with a double urinary stream (the most common presentation), urinary tract infection, incontinence, epididymitis, and bladder outflow obstruction. The authors have not described the type of duplication very well and the reader will be confused whether it is a complete, Y-type or some other type of duplication. Most pediatric urologists and surgeons agree upon identifying the functional urethra prior to any surgical correction of this anomaly. The functional urethra can be determined by urodynamic studies of both urethrae but it was not done in this study. I humbly appreciate the authors performance of cauterization of the ventral urethra; however, the remnant of the ventral urethra is observable on post-operative MCUG. This remnant is concerning for its potential to cause urinary tract infection or incontinence in the future.

**I have read this submission. I believe that I have an appropriate level of expertise to state that I do not consider it to be of an acceptable scientific standard, for reasons outlined above.**

**Competing Interests:** No competing interests were disclosed.

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