



## Complete duplication of the penis - A case report

Saidanvar Agzamkhodjayev<sup>a,b</sup>, Kobiljon Ergashev<sup>a,\*</sup>, Zafar Abdullayev<sup>a,b</sup>, Asqar Soliyev<sup>a</sup>, Ruslan Batrutdinov<sup>c</sup>

<sup>a</sup> Department of Urology, National Children's Medical Center, Tashkent, Uzbekistan

<sup>b</sup> Department of Urology, Tashkent Pediatric Medical Institute, Tashkent, Uzbekistan

<sup>c</sup> Scandinavia Clinic, St Petersburg, Russian Federation

### ARTICLE INFO

#### Keywords:

Penile duplication  
Diphallia  
Congenital anomaly

### ABSTRACT

Penile duplication is a very rare urological entity. It may be associated with other congenital conditions such as urogenital, GI tract and musculoskeletal anomalies. Properly classifying the condition may dictate the final treatment options. Our current case is the complete true duplication in which we performed side-to-side urethra-urethral anastomosis. We spared the posterior urethra as it may end up with postoperative urinary incontinence.

### 1. Introduction

Duplication of the penis is a rare anomaly with the incidence of 1 in 5 or 6 million. It may be presented as isolated or in part with other congenital urogenital anomalies as ectopic or horseshoe kidney and bladder duplication, GI tract abnormalities as imperforate anus, and also problems with musculoskeletal field as pubic diastasis and lumbosacral anomalies.<sup>1</sup> The index case was also associated with anal atresia which was operated previously right after birth. Although each penis is structurally normal, it is a great surgical challenge for the surgeon. In this paper we report a case of complete penile duplication and its surgical management in a 7-year-old boy.

### 2. Case report

A 7-year-old male child was referred to our clinic with abnormal genitalia. Initial physical examination identified two completely developed penises with common shaft skin (Fig. 1A). He was fully continent and there was a urine flow from both urethras. On his first day of life, proctoplasty was performed to fix the anal atresia. MRI (Fig. 1B) showed two completely developed penises with each having separate two cavernous bodies, one spongy body. Abdominal USG and CT urography showed normal KUB on each side with an aberrant vessel to the right kidney. Voiding cystourethrogram (VCUG) showed two separate urethras opening into the single bladder via a single bladder neck without any signs of reflux. Uroflowmetry studies were performed

through right and left penises separately with normal  $Q_{max}$  - 11.8 and 10.3 ml/s, respectively.

Preoperatively, retrograde urethrogram (Fig. 1C) and cystourethroscopy were performed through both urethrae and VCUG findings were confirmed. Interestingly, verumontanum was common for both urethras (Video 1). We performed left partial penectomy (Fig. 2A), side-to-side urethra-urethral anastomosis in anterior urethra. The latter was achieved between spatulation of the proximal end of the left penile urethra and the side of the proximal part of the right penile urethra. Two catheters were placed: one into the bladder to drain the urine postoperatively (percutaneous cystostomy) (Fig. 2B); second through the right urethra and across the anastomosis. Urethral catheter was removed on the 10th and cystostomy catheter was removed on the 21st postoperative days.

Supplementary video related to this article can be found at <https://doi.org/10.1016/j.eucr.2021.101892>

The patient was fully continent with normal voiding and postoperative two months uroflow study was performed revealing normal  $Q_{max}$  of 14.2 ml/s (Fig. 2C). Informed consent was obtained from the parents of the patient.

### 3. Discussion

Penile duplication is a rare congenital anomaly with the incidence of 1 in 5–6 million. Although the anomaly is known from the XVII century, there are only just over 100 cases reported so far.

*Abbreviations:* Ultrasonography of the kidneys ureters and bladder, (USG KUB).

\* Corresponding author. Department of Urology, National Children's Medical Center, Tashkent, Yashnabad district, Parkent street, 294, Uzbekistan.

E-mail address: [kobiljonergashev@gmail.com](mailto:kobiljonergashev@gmail.com) (K. Ergashev).

<https://doi.org/10.1016/j.eucr.2021.101892>

Received 9 September 2021; Received in revised form 5 October 2021; Accepted 11 October 2021

Available online 14 October 2021

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Fig. 1A. Two completely normally developed penises with a common shaft.

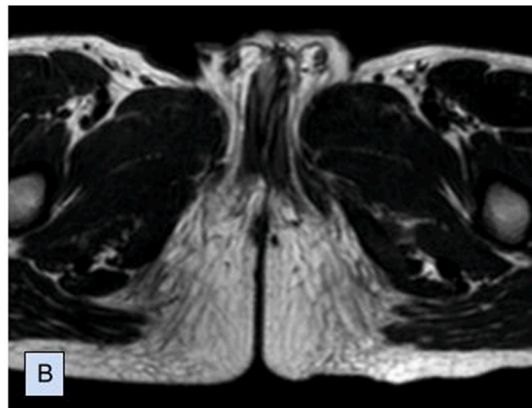


Fig. 1B. MRI study of the pelvic organs including penile structures.

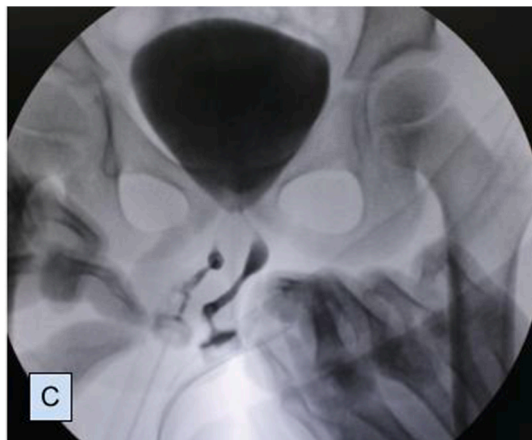


Fig. 1C. Intraoperative retrograde urethrogram.

Most of the penile duplication cases are associated with urogenital, anorectal malformations and musculoskeletal anomalies. Associated urogenital anomalies include bladder exstrophy, bladder duplication, bifid scrotum and renal anomalies.<sup>1</sup>

Key mechanism behind the embryogenesis of penile duplication is indefinite, but can be explained with: (1) “separation” of the pubic tubercles during embryogenesis, in which each phallus has one corporal body and urethra, or (2) “cleavage” of the pubic tubercle in which each

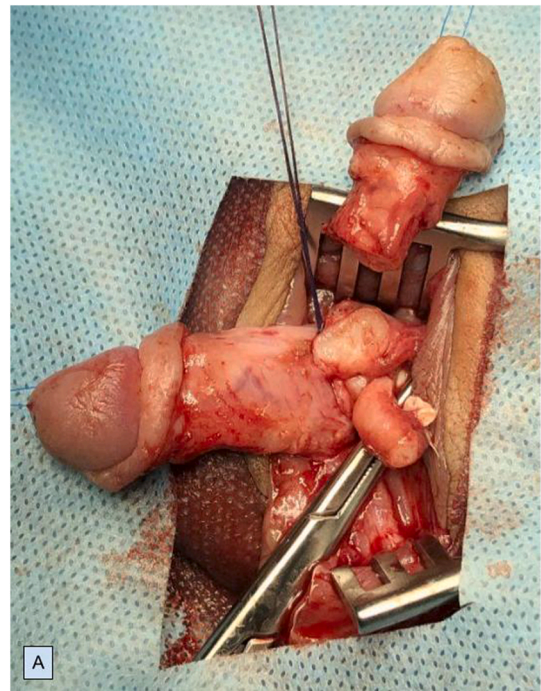


Fig. 2A. Partial left penectomy.



Fig. 2B. Two catheters: urethral and cystostomy tubes.



Fig. 2C. Postoperative two months appearance.

phallus has 2 corporal cavernous bodies and urethras.<sup>2</sup>

Gyftopoulos et al.<sup>3</sup> proposed a classification where cases can be divided into 2 broad categories: true diphallia and bifid phallus. Both can be subclassified into partial or complete duplication. True complete diphallia will have 2 well-developed penises (with 2 corpora cavernosa and 1 corpora spongiosum).<sup>4</sup> True partial diphallia will have a smaller or rudimentary duplicate penis (with complete structures that is 2 corpora cavernosa and 1 corpora spongiosum). If the duplicate penis does not have all the structures, for example one corpora cavernosum they are classified as bifid phallus. Depending on the degree of separation, bifid phallus is further subclassified into complete and partial. Complete bifid phallus has separation at the base whereas, partial bifid phallus has separation at the glans.

According to Gyftopoulos' classification, our case has true complete diphallia.

We performed side-to-side anastomosis between anterior parts of urethras, in the proximal penile part. This resulted in structural and functional urethra. In order to maintain the continence postoperatively, our dissection did not include the posterior urethra.

Diphallia is a rare congenital anomaly that can be associated with urogenital, anorectal malformations. Hence the presentation of the patients may be different and requires an individualized approach. Side-to-side urethral anastomosis with avoiding dissection in the posterior

urethra results in a satisfactory outcome.

#### Authors contribution

KE: Conceptualization, Investigation, Resources, Writing - Original Draft.

ZA: Conceptualization, Methodology, Data Curation, Supervision.

SA: Supervision. All authors have read and approved the final manuscript.

#### Declaration of competing interest

The authors declare no conflicts of interest.

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