OPEN ACCESS

PAKISTAN JOURNAL OF UROLOGY

Pakistan J Urol
2024, 02(1): P:118-122

ISSN: 3005-7582 (Online) : ISSN: 3005-7574 (Print)

A CASE SERIES

Diphallia A Unique Case Series Of A Rare Congenital Anomaly

Sunirmal Choudhury[1], Sinha Subham[2], Katiyar Vipin[3], Gopalakrishna- Rk[4], Patel Prakhar[5]

[1,2,3,4,5-]Department Of Urology, Kolkata Medical College, India

ABSTRACT

INTRODUCTION

Diphallia, or double penis, is very rare, and there are very few reported cases in the literature. The incidence is 1 in every 5 to 6 million live births. [1] Approximately 100 cases have been reported since the first case reported by Wecker in 1609.[2] The extent of duplication and the number of associated anomalies vary greatly, ranging from a double glans arising from a common shaft with no other anomaly to complete duplication of the phallus accompanied by multiple anomalies.[3] Embryologically, a diphallus deformity arises from either "separation" of the pubic tubercle, wherein each phallus will have only one corporal body and urethra, or "cleavage" of the pubic tubercle, where each phallus will have two corporal cavernous bodies and urethras. [4, 5] Diphallus has been classified in different ways, such as glandular, bifid, concealed, complete, hemi-diphallus and triple penis. [5.6] The majority have a single corpus cavernosum in each organ. [7] We hereby report a case of a double penis and associated multiple congenital abnormalities.

Keywords: Diphallia, Congenital anomaly, Embryology, Duplication, Multiple abnormalities

CASE REPORT 1:

A 14-year-old boy came to our outpatient department with complaints of pain and swelling over his left scrotum, associated with fever for the last 6 days. There is a history of multiple surgeries.one in the neonatal period, where colostomy was made for the imperforate anus (no documentation), and another in early childhood (2013), where the closure of the rectovesical fistula and vesico-vesicostomy was performed. On local examination, there was a left scrotal abscess, and the patient had a double phallus. On physical examination, the right phallus is of normal size and was placed laterally with the external urethral meatus in a normal position. The left phallus is smaller, with epispadias meatus located at the midline. [FIG 1] His left scrotum is well developed with normal testis,the right scrotum is underdeveloped, and the right testis is located in the right inguinal region. [FIG 2a] On examination, we also found that the patient had an imperforate anus for which a colostomy was made during the early neonatal period.

How to Cite : **Khan, R. A., Khan, Z. A., & Fahad, S. (2024). Demographics and Clinical Outcomes of Hyperkalemia in Hospitalized Patients: A Detailed Examination: Original Article. Pakistan Journal of Urology (PJU), 2(01), 118–122. https://doi.org/10.69885/pju.v2i01.54**

Corresponding Author: Sunirmal Choudhury

Department Of Urology, Kolkata Medical College, India

Email: sunir09@gmail.com

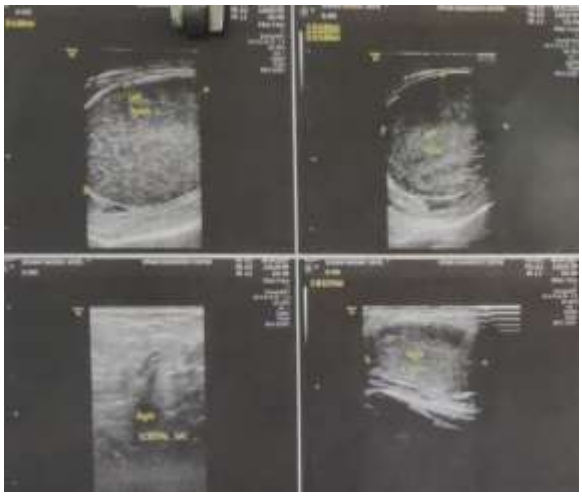
ORCID ID: 0000-0002-1899-9196

Article History

Received:	January	05-2024
Revision:	February	22-2024
Accepted:	April	17-2024
Published:	July	05- 2024

Figure 1: Showing Double Phallus With Right-Sided Under Developed Scrotum**.**Presence Of Fat Pad Above Right Phallus

Figure 1: Fig 2a: Right Undescended Testis Identified In Right Inguinal Region



On further evaluation: USG of the lower abdomen & penis was done, which showed two bladder cavities with an incomplete septum and two phalli with both having two corpora cavernosa each- suggestive of complete diphallia. [FIG 2b&2c]

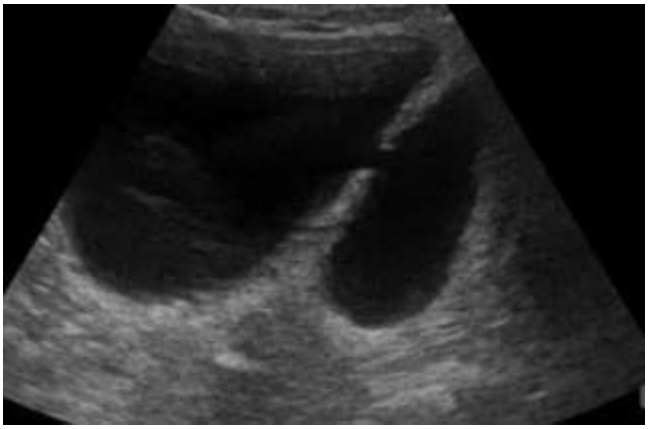


Fig 2b: two bladder cavities showing communication with each other**.**

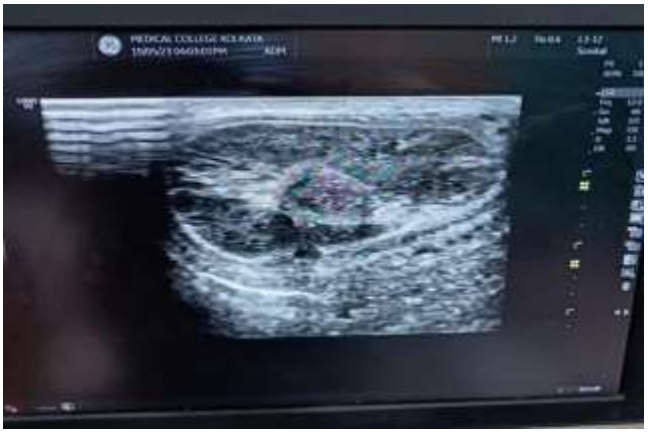


Fig 2c: Urethra With 2 Corpora Cavernosa

RETROGRADE URETHROGRAM suggested two separate urethra with two bladder cavities, the left of which has a smaller urethra, smaller bladder cavity and spatulous bladder neck. The right phallus appears normal. There is communication between bladder cavities. [FIG 3]

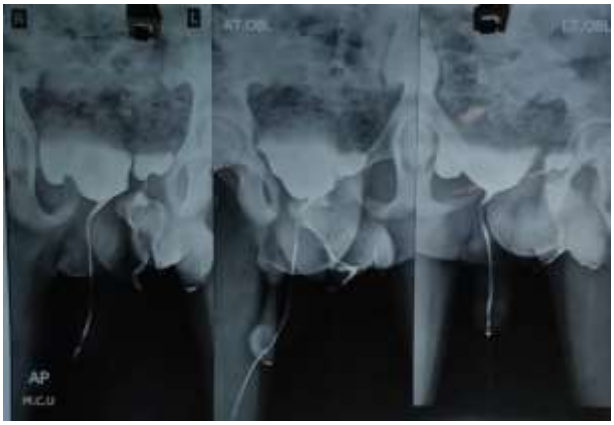


Figure 3 shows a double bladder cavity with communication with a normal right urethra.

X-ray of the axial skeleton shows severe kyphoscoliosis with pubic diastasis.

A whole CECT abdomen with oral contrast was done to identify other congenital abnormalities, which showed the presence of a normal left kidney with an absent right kidney. We took the patient for cystoscopy under anaesthesia. Cystoscopy through the right phallus showed a normal urethra and bladder neck, with a normal capacity for the bladder cavity. A single ureteric orifice was seen in this cavity. Cystoscopy through the left phallus showed an underdeveloped bladder neck with a small bladder cavity, and communication with the right bladder cavity could be identified [FIG 6a&6b]

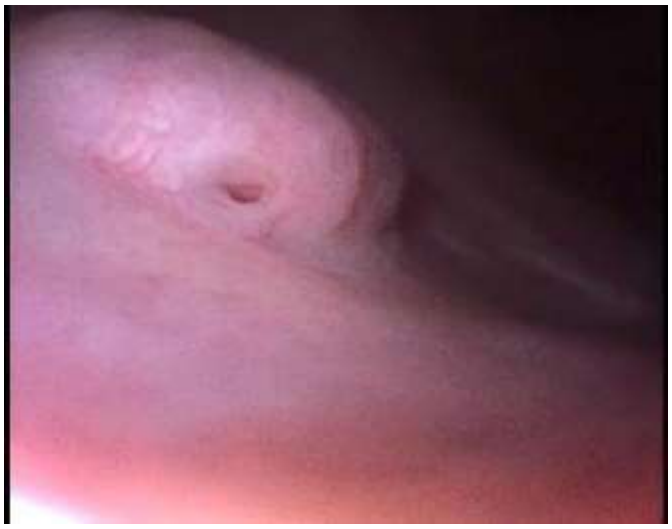


Figure 4a: single ureteric orifice

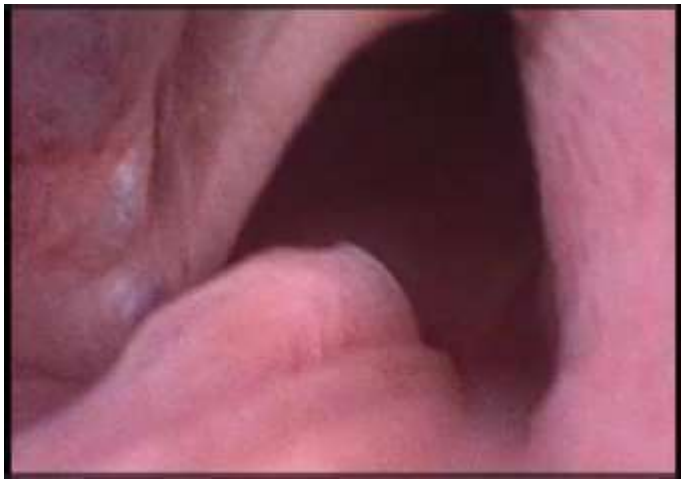


Figure 4b: Communication Between Both Cavities

We initially managed the patient with emergent drainage of scrotal abscess and control of septic features. Then, we evaluated the patient for definitive management of double phallus. After consultation with other disciplines (paediatric surgery, plastic surgery, psychiatry, endocrinology), we have planned for reconstructive surgery after the patient attains adulthood (as per psychiatry opinion).

CASE REPORT 2:

A 63-year-old male patient presented with left-sided scrotal pain, swelling and fever in May 2013 in a tertiary care hospital in the eastern part of India. The patient also had a history of urge incontinence.

On local examination, there was swelling and redness of the left testis. The cord structures were also found to be inflamed. Clinically, he was diagnosed with a case of left-sided epididymorchitis. One interesting finding was noted during local examination – he had two penile shafts, both of which were of equal length, with external urethral meatus located in a normal position. Both of those phallus had erectile function, and the patient was completely continent. He also had a normal sexual life and had two healthy children. [FIG 5]

Figure 5: Double Phallus With Left Epididymo Orchitis

On investigation, USG (inguinoscrotal region) showed that the left kidney was non-visualized, with right-sided compensatory hydronephrosis and left-sided minimal hydrocoele. This patient was initially managed with conservative treatment, and his symptoms were resolved. We could not evaluate the patient any further as he did not seek any treatment and was lost during follow-up.

DISCUSSION:

Schneider classified Diphallus into three groups: diphallia of glans alone, bifid diphallus & complete diphallia. [6] Vilanova and Raventos have recently added a fourth category called pseudodiphallia.[8] Recently accepted classification defines the following two major groups: true diphallia and bifid phallus. These two groups are further divided into partial or complete duplication. True complete diphallia is characterized by complete penile duplication, with each phallus having two corpora cavernosa and one corpus spongiosum. If the duplicate penis is smaller or rudimentary with complete structures, it is described as a true partial diphallia. When there is only one corpus cavernosum in each penis, the term bifid phallus is used. Moreover, if the degree of separation is complete to the base of the shaft or just to the glans, the anomaly is described as complete or partial bifid phallus, respectively. [13] Our case can be considered as a case of true complete diphallia as each phallus showed the presence of two corpora cavernosa and one corpus spongiosa. True diphallia is the less common variety and usually presents with a wider range of associated malformations such as bladder and urethral duplication, exstrophy vesica, renal anomalies, bifid scrotum, anorectal malformations, bowel duplication, and vertebral anomalies.[13,14]. Bifid phallus is usually associated with less severe malformations.[13] True diphallia can rarely be isolated.[15] Muramatsu et al. [16] reported a case of a 15-year-old patient presenting with VATER syndrome, chronic renal failure, and penile duplication, including a hypoplastic lower urinary tract. In our case, there was bladder duplication along with multiple congenital anomalies.The meatus may be normal, hypospadiac, or epispadiac. The scrotum may be normal or bifid. Various studies have reported several associated congenital anomalies such as bifid scrotum, hypospadias, duplicated bladder, imperforate anus, bladder exostrophy, colon duplication, inguinal hernia and renal agenesis.[9-12] It may be associated with other malformations like cloacal anomaly, colon duplication, urethral duplication, a horseshoe kidney, undescended testes, a hypoplastic right leg, and a ventricular septum defect. [13] In our case, episodic meatus, right undescended testis, single left kidney, imperforate anus and skeletal abnormalities were noted. Penile duplication and multiple associated anomalies can be managed with multiple surgeries (treatment of the associated anomalies and genitalreconstruction).[14] De Oliveira et al. reported an isolated true partial diphallia that was managed in a single surgery with right penectomy and end-to-side anastomosis between the right urethra and the remaining urethra. [15] Excision of the ventral penis and using its preputial skin for hypospadias repair (Duckett tube) of the dorsal penis has also been reported in the literature.[1] Another case with imperforate anus, double bladder, and duplicated penis, reported by Mirshemirani et al., was managed with laparotomy and colostomy on the third day of life, and cystoplasty and reimplantation of the left ureter in a single bladder and resection of left phallus were performed when the patient was 4 months old. [12] If there is one corpus cavernosum in each penis, joining two corporal bodies with penile reconstruction is preferred. [17,18] Elsawy et al.reported a joining technique without the removal of any penis, even for true diphallia with two corpora in each penis. [19]

CONCLUSION:

Penile duplication is a rare, common congenital anomaly. Systematic investigations are mandatory in all cases to expose essential congenital malformations that are theoretically life-threatening and require immediate surgical correction. This has a profound psychological impact on the entire family and the child in particular; decisions have to be taken early and should always be personalized according to the amount of penile duplication and the degree of the associated anomalies. All patients with penile duplication have to be evaluated carefully because of the high incidence of other systemic anomalies, and all can be repaired surgically. A multidisciplinary approach should be taken with the individualization of each case.

Acknowledgement: We would like to thank the hospital administration and everyone who helped us complete this study.

Disclaimer: Nil **Conflict of Interest:** Nil **Funding Disclosure**: Ni

Authors Contribution:

Concept & Design of Study: Sunirmal Choudhury1

Drafting: Sinha Subham2, Katiyar Vipin3

Data Analysis: , Gopalakrishna- Rk4, Patel Prakhar5

Critical Review: Gopalakrishna- Rk4, Patel Prakhar5

Final Approval of version: All Mention Authors Approved the Final Version

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