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Case Report

True partial diphallia with associated penoscrotal transposition of two hemi-scrotums ^{☆,☆☆}

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ABSTRACT

Diphallia or duplication of penis is extremely rare condition with a reported incidence of 1 in 5-6 million live births. Approximately around 100 cases of diphallia have been described in literature, each case have a unique presentation from associated anomalies. Clinically these patients can be classified into complete (true diphallia) or partial duplication. In true diphallia, each penis has 2 corpora cavernosa and 1 corpus spongiosum. If the duplicate penis is smaller or rudimentary with complete structure, it is described as true partial diphallia. The term bifid phallus is used if there is only one corpus cavernosum in each penis. Due to low incidence and varied presentation, not much is known about the underlying pathophysiology, management options, and outcomes. Here, we report a case of partial diphallia with associated penoscrotal transposition of 2 hemi-scrotums.

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Background

Diphallia or duplication of penis is extremely rare condition with an estimated incidence of 1 in 5-6 million live births [1,2]. Approximately about 100 cases have been described in the literature, each case have a unique presentation, from associ-

ated various anomalies. The reported anomalies include duplication of bladder, urethra, and/or colon, anorectal malformations and vertebral anomalies [1-3].

There is no universally accepted classification system, but the cases can be broadly classified into true diphallia or bifid phallus. In true diphallia each penis has 2 corpora cavernosa and 1 spongiosum. If there is partial duplication, that is if the

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Fig. 1 – Physical examination demonstrated 2 completely developed penis with normal looking meatus, both penises covered at the mid-part of the shaft with one penile skin shaft and penoscrotal malposition.

penis has one corpora cavernosum in each penis, it is classified as bifid phallus [1,2,4].

Due to low incidence and varied presentation, not much is known about the underlying pathophysiology, management options and outcomes [2,5–7]. Here, we report a case of diphallia with associated penoscrotal transposition of 2 hemiscrotums.

Case report

A 6-month-old full-term infant presented to the urology clinic for management of diphallia (Fig. 1). No reported relevant past medical history. Physical examination demonstrated 2 com-

pletely developed penis with normal looking meatus, both penises covered at the mid-part of the shaft with one penile skin shaft, penoscrotal malposition, right side scrotal skin tag, sacral dimple, and undescended testes. Both of them have urine flow from the urethras. Amniotic karyotype was 46 XY.

Ultrasound of the penis and scrotum (Fig. 2) demonstrated well-developed right penis, with normal corpora spongiosum and normal separate corpora cavernosa each supplied by separate cavernosal artery. Left penis demonstrated normal corpora spongiosum, and 2 separate corpora cavernosa supplied by 1 cavernosal artery. Undescended well-developed testes were also seen in the inguinal canal (Fig. 3). Doppler imaging demonstrated normal flow to both the testes.

Vesicocystourethrogram (Fig. 4) demonstrates normal flow during voiding phase in the well-developed right penile urethra. The left penile urethra was relatively smaller in caliber (but patent) and proximally it terminated in the right prostatic urethra. The imaging findings were consistent with true partial diphallia.

The patient is scheduled for resection of left penis at the level of the base of left penile urethra.

Discussion

The first case of diphallia was reported in 1609 [4,8]. Following which approximately 100 cases have been reported in the literature with varied presentations. These cases are associated with other urogenital and anorectal malformations, such as hypospadias, epispadias, exstrophy bladder, duplication of bladder, renal agenesis, caudal duplication syndrome, imperforate anus, duplication, and triplication of the colon [1–4].

Underlying mechanism is unclear, but postulated to be: (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 phallus has 2 corporal cavernous bodies and urethras [4].

Gyftopoulos et al [4] proposed classification where cases can be divided into 2 broad categories: True diphallia and bifid

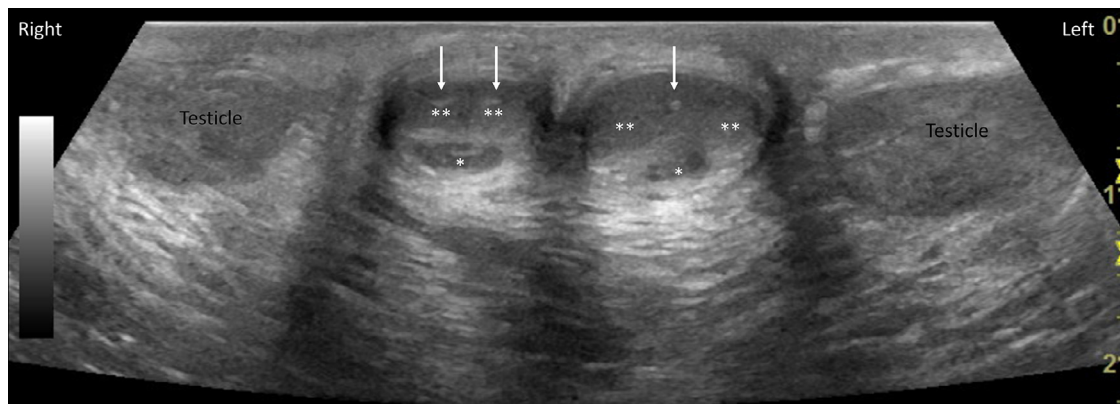


Fig. 2 – Ultrasound of the penis and scrotum demonstrates well-developed right penis, with normal corpora spongiosum (*) and normal separate corpora cavernosa () each supplied by separate cavernosal artery (white arrow). Left penis demonstrated normal corpora spongiosum (*), and 2 separate corpora cavernosa (**) supplied by one cavernosal artery (white arrow).**

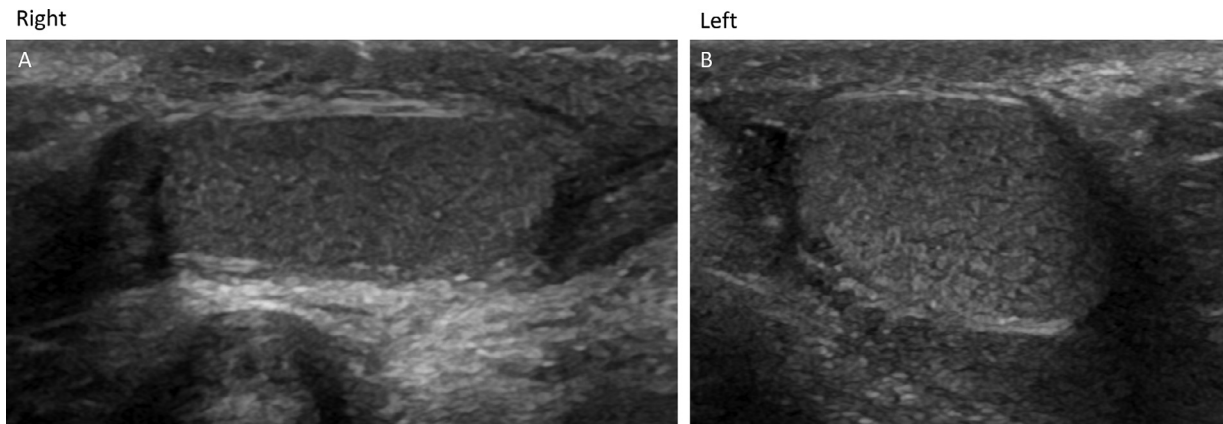


Fig. 3 – Undescended well-developed testes noted in the inguinal canal. Doppler imaging demonstrated normal flow to both the testes (not shown in the figure).



Fig. 4 – Voiding phase of the vesicocystourethrogram demonstrates a well-developed urethra of the right penile shaft (white arrow). The left penile urethra is relatively smaller in caliber (but patent), which proximally terminates to the main prostatic urethra (yellow arrow).

phallus. Both can be subclassified into partial or complete duplication. True complete diphallia will have 2 well-developed penis (with 2 corpora cavernosa and 1 corpora spongiosum). True partial diphallia will have 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 [4].

Due to wide variability in presentation the management strategies differ [5–7,9]. Surgical correction is individualized with the aim of achieving proper urinary continence and erection with adequate cosmesis [10,11].

Karagözlü et al [12] recommend complete penectomy and urethral-urethroostomy while preserving the posterior

urethra may be a better choice for individualized treatment of diphallia, without the risk of damaging the sphincter and the prostate gland.

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