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True Diphallia: A Report of Two Cases

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Authors' contributions

This work was carried out in collaboration among all authors. Author MCA designed the study. Author AG performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors AS and VV managed the analyses of the study. Authors RS and MS managed the literature searches. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aims: Diphallia or penile duplication is an extremely rare congenital anomaly. It occurs once in every 5.5 million live births. Wecker (1609) reported the first case in Bologna, Italy. It varies from a small accessory penis or duplication of the glans to complete penile duplication.

Presentation of Case: A 31 years old man presented to with urinary incontinence since birth. Examination revealed skin covered exstrophy with two phalli lying side by side in sagittal plane.

Case 2: A 3 month old infant was brought to us with complete duplication of penis and urethra, two phalli lying side by side and voiding from both. Both the testes were normally descended and anal opening was normal.

Discussion and Conclusion: Penile duplication is rare and no two cases are identical. It can be true complete diphallia, partial diphallia, complete bifid phallus or partial bifid phallus. The meatus may be normal, hypospadiac, or epispadiac and the scrotum may be normal or bifid. The testes may be normal, atrophic, or undescended.

Treatment aims at preserving continence, erectile function and aesthetically acceptable genitalia.

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Keywords: Diphallia, bifid phallus; mitrofanoff; hypospadiac meatus.

1. INTRODUCTION

Diphallia or penile duplication is an extremely rare congenital anomaly. It occurs once in every 5.5 million live births [1]. Wecker reported the first case in Bologna, Italy [2] Smail acimi [3] reviewed the literature and found 93 such cases. It varies from a small accessory penis or duplication of the glans to complete penile duplication. Herein, we report 2 cases of penile duplication with review of literature.

2. CASE REPORTS

2.1 Case 1

A 31 years old man presented to us with urinary incontinence since birth. Examination revealed skin covered exstrophy with 2 phalli lying side by side in sagittal plane. There was no urethral plate in either phallus with penopubic epispadiac meatus. Testes, anus and spine were normal. Divarication of recti was present. Erectile status was poor in both phalli. He denied any history of masturbation or ejaculation. On examination, each phallus was having two corporeal bodies further confirmed by ultrasound. Complete blood counts (CBC), Serum Creatinine and Random blood sugar were normal. X-ray KUB showed pubic diastasis. Intravenous urogram (IVU) depicted normal bilateral upper tract with adequate bladder capacity (Fig. 2). In view of absent urethral plate, shared decision was taken to remove 1 phallus with bladder neck closure and a diversion using Mitrofanoff's procedure (appendicovesicostomy). Post operative period was uneventful and he is on regular clean intermittent catheterisation (CIC).

2.2 Case 2

A 3 month old infant was brought to us with complete duplication of penis and urethra, two phalli lying side by side and voiding from both (Fig. 3). Both the testes were normally descended and anal opening was normal. His parents were advised for admission and further evaluation. However, they were reluctant and lost to follow-up.

3. DISCUSSION

Penile duplication is rare and no two cases are identical. Neugebauer reviewed 28 cases of penile duplication (among 37 cases of external genitalia duplication) in 1898 (quoted by Nesbit and Brommein 1933) [4] In 1950, Pendino surveyed 53 cases [5] Till date about 100 cases have been reported.

Aleem et al [6] divided diphallia into four groups:

1. **True complete diphallia:** Complete penile duplication, each with two corpora cavernosa and a corpus spongiosum.
2. **Partial diphallia:** The duplicate penis is smaller or rudimentary.
3. **Complete bifid phallus:** When only one corpus cavernosum is present in each penis upto base of shaft.
4. **Partial bifid phallus:** When degree of separation is not complete to the base of shaft.

The meatus may be normal, hypospadiac, or epispadiac and the scrotum may be normal or bifid. The testes may be normal, atrophic, or undescended.

Penis normally develops with fusion of bilateral cloacal tubercles; failure of this might lead to bladder exstrophy/epispadias with a split phallus in place of true diphallia. Complete penile duplication is explained embryologically by the possibility of a longitudinal duplication of cloacal membrane finally resulting into two genital tubercles. Diphallia is usually associated with other malformations, such as bladder and urethra duplication, exstrophy of the cloaca, exstrophy of the bladder, anorectal malformation, colon and rectosigmoid duplication, ventral hernia, pubic symphysis diastasis, abnormality of skeletal and heart muscles, and vertebral anomalies [7,8]. The theory of duplication of cloacal membrane also explains associated bladder, ano rectal and spinal anomalies. Karyotype analysis depicts chromosomal balanced reciprocal translocation 46, XY, t(1,14)(p36.3; q24.3) was reported by kapur etal and implicated defects in the homeobox genes in diphallia formation [9,10,11].

The degree of erectile function in cases of diphallia varies significantly. Usually, one or both penises have erection. In cases of true, complete diphallia presented at advanced age, simultaneous erection and, occasionally, ejaculation have been reported. Ultrasound can be used to detect the number of corpora or

Fig. 1. Skin covered exstrophy, two phalli with penopubic meatus

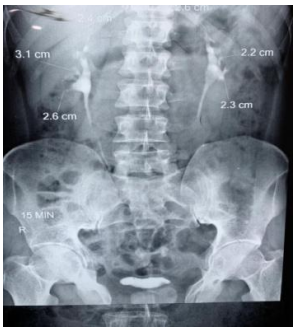

Fig. 2. IVU shows bilateral upper urinary tract and pubic diastasis

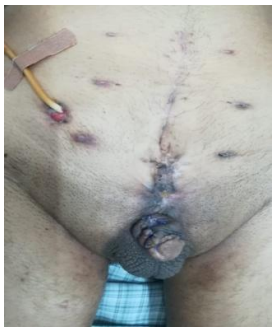

Fig. 3. Post operative clinical photograph

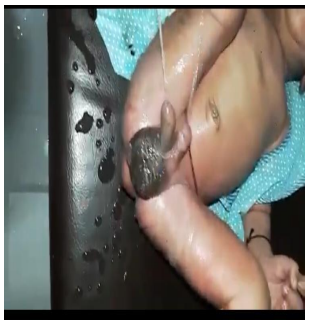

Fig. 4. 3 month old infant with complete diphallia

corpus spongiosum. Magnetic resonance imaging (MRI) better delineates anatomic abnormalities and can be used before contemplating surgical intervention.

Treatment aims at preserving continence, erectile function and aesthetically acceptable genitalia. It depends on the type of accompanying congenital abnormalities. In cases of bifid phallus, an endeavour should be made to dissect and join them together to restore normal anatomy. Nevertheless, in cases of true diphallia, excision of redundant or extra phallus is needed. Priyadarshi and Djordjevic et al. reported penis reconstruction by joining the corporal bodies in each penis in a patient with true complete diphallia [8,12,13].

4. CONCLUSION

Penile duplication is rare and no two cases are identical. It can be true complete diphallia, partial diphallia, complete bifid phallus or partial bifid phallus. The meatus may be normal, hypospadiac, or epispadiac and the scrotum may be normal or bifid. The testes may be normal, atrophic, or undescended.

Treatment aims at preserving continence, erectile function and aesthetically acceptable genitalia.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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