

## CASE REPORT

### DIPHALLIA: REPORT OF A CASE

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#### ABSTRACT

**We report on a case of penile duplication in a thirteen-year-old African boy. The diphallia was incomplete. There was a common root of the penis with accessory glans and a urethra in a hypospadias position under a common prepuce. Only one urethra left the bladder along the left penile shaft and ended in a normally positioned meatus. There were no other associated congenital anomalies. After surgical repair, the patient had a good functional and cosmetic result. In the light of our case and the review of the literature, the classification, associated anomalies and the aetiology of diphallia are discussed.**

#### INTRODUCTION

Diphallia is a rare congenital condition, estimated to occur once in 5.5 million births in the United States<sup>1</sup>. The first case of diphallia was reported in 1609 by Wecker<sup>2</sup>. Since 1993, cases of diphallia have been reported in a rate of about 1 every 2 years<sup>3</sup>. During the last 10 years, there has been a noticeable and unexplainable increase in reports on diphallia which might be due to a growing awareness of this condition.

Diphallia presents in various ways. The extent of diphallia and the number of accompanied anomalies vary greatly ranging from rudimentary erectile tissues with no other anomaly to a complete true duplication of the phallus associated with variable anomalies. Anomalies are to be expected; they occur at random, many are clustered, and they should be sought out and treated as early in the patient's life as possible because they are the principal cause of mortality in these individuals.

We herein report our experience with the first case of diphallia in our center and discuss the classification and proposed embryogenesis of diphallus.

#### CASE REPORT

A 13-year-old African child was referred to our Department of Urology presenting with dysuria and expressing his unhappiness about the configuration of his penis. Otherwise, he was a healthy boy with a normal birth history. Both his parents and brothers were normal.

On examination, the boy was found to be in a good general condition. Physical examination revealed no obvious anomaly except for diphallia. Examination of his perineum did not reveal any anal anomaly. On genital examination a normal scrotal development was noted; the testes were located in the scrotum and were of normal size, shape and consistency. He had one phallus with normal length and shape located in the midline.

On further examination, the median raphe was noted to be displaced to the phallus (Fig. 1). Retraction of the prepuce revealed an anti-clockwise rotation of the glans with a normally positioned urethral meatus (Fig. 2). On the right side of the glans there was an extra glandular tissue with a patent, but hypospadias urethra. On catheterization, the urethra extended proximally on the shaft by about 10 cm



Fig. 1: Preoperative picture showing deviation of the median raphe

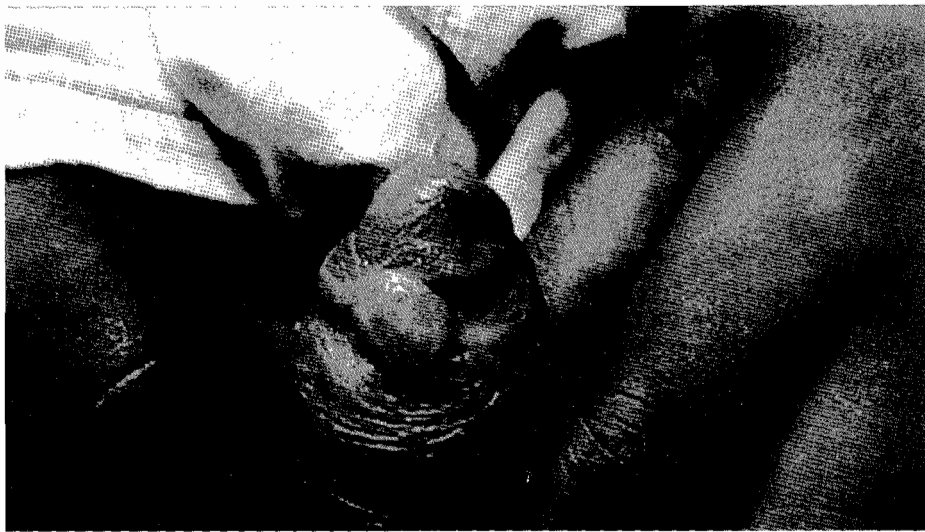


Fig 2: Preoperative picture demonstrating the malrotated glans and accessory glans on the right side with hypospadias urethra (not shown in the picture)

(Fig. 2). The boy passed urine through the left urethral meatus on the tip of the normal appearing glans.

Laboratory tests included blood chemistry and urine analysis. Urea and creatinine were found to be normal.

Ultrasound examination of the abdomen showed no upper urinary tract anomaly.

A voiding cystogram was done and showed a normal bladder with no signs of a septum within the bladder and a normal single urethra connected to the bladder with no residual voiding.

Intraoperatively, the right urethra was cannulated. The right phallus extended to about 10 cm and the accompanied urethra ended blindly. The right glans with the associated urethra



Fig. 3: Postoperative picture taken one week after the operation showing a satisfactory cosmetic result

was mobilized, freed from the rest of the phallus and excised at the most proximal part. The skin was then approximated and closed to cover the defect.

The postoperative period was uneventful. Due to lack of experience with such anomaly, the rotation of the penis was not completely corrected, however the patient was satisfied with the cosmetic result (Fig. 3).

## DISCUSSION

Diphallia is a rare urogenital anomaly occurring in about 1 in every 5 million live births<sup>1</sup>. Due to the rarity of the condition, its incidence within different racial groups has not been documented. According to the literature, diphallia presents in many different ways. The main variation is the presence or absence of associated anomalies as well as the extent of such anomalies which may range from fusion of the two phallia to their complete separation.

### Classification:

The classification after Abdel Aleem<sup>4</sup> is the most widely accepted classification of diphallia<sup>4</sup>. It differentiates between true diphallia and bifid phallus.

True diphallia may be complete (the accessory organ has 2 corpora cavernosa and 1 corpus spongiosum) or partial (the accessory

organ is only rudimentary and presents as a glans or small piece of erectile tissue).

A bifid phallus may be partial presenting a bifid glans or a bifid shaft or it may be complete.

### Associated Anomalies

After reviewing the literature, the authors would like to suggest an amendment of the classification of associated anomalies into genitourinary, gastrointestinal and musculo-skeletal anomalies as follows:

#### A: Genitourinary Anomalies

##### 1. Urethra

- \* absent urethra in one or both phalli
- \* double urethra complete or incomplete
- \* hypospadias or epispadias

##### 2. Bladder

- \* extrophy alone
- \* extrophy with vesico-intestinal fissure
- \* bladder duplication, complete/incomplete

##### 3. Scrotum

- \* scrotal left
- \* bifid scrotum

**B: Gastrointestinal Anomalies**

- \* imperforate anus with or without recto-urinary fistula
- \* duplication of the colon

**C: Musculoskeletal Anomalies**

- \* Diastases of the pubic bones without bladder extrophy
- \* Lumbosacral anomalies
- \* Clubfoot
- \* Hernia of the lower abdominal wall

**Aetiology:**

The aetiology of the diphallia is still hypothetical and not well understood. It is difficult to explain the duplication of the penis, since at no time in normal development is the genital tubercle a paired structure.

Rodriguez (1965)<sup>5</sup> suggested that diphallia was due to the lack of fusion of the paired mesodermal anlagen of the genital tubercle, while Campbell (1951)<sup>1</sup> assumes that duplication of the bladder and penis may be caused by splitting of the vesico-urethral anlagen. Another theory was introduced by Satter and Mossman (1958)<sup>6</sup>; they believe that the aetiological factor for the duplication of the urogenital organs might be a doubling of the

original endodermal allantoic anlagen. On the other hand, Abdel Aleem (1972)<sup>4</sup> suggests that an early separation of the pubic bones may cause a deviation of the embryonal cavernous tissue arising from both ischiopubic synostoses. When this cavernous tissue reaches the surface, a phallus may develop.

We conclude that due to the rarity of the phenomenon some extensive research is still required to clarify the actual aetiology of diphallia.

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