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Case Report

Rare case of isolated true complete diphallus – Case report and review of literature

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Abstract: Penile duplication is very rare anomaly. True complete diphallia is mostly associated with severe anomalies. Isolated complete diphallia is extremely rare. The case is being presented as true penile diphallia without associated anomalies. We are discussing diagnosis and management of such rare cases.
Keywords: penile duplication, true diphallia, completes diphallia

INTRODUCTION

Diphallia or penile duplication is an extremely rare congenital anomaly. It occurs in every 5.5 million live births [1]. The extent of duplication and 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 such as ectopic scrotum, bifid scrotum, hypospadias, imperforate anus, bladder extrophy, colon duplication, double bladder and vertebral anomalies [2]. Isolated complete diphallus is extremely rare [3, 4, 5]. Most of isolated cases had hypospadiac meatus or rudimentary phallus but in our case it was complete duplication with well developed corpora cavernosa and spongiosum in both phalluses. Case is being presented due to rarity of this condition.

CASE REPORT

A 10 year old boy presented with duplication of penis [Fig 1]. He had normal scrotum, two descended testicles and patent normal positioned anus. The right sided penis was smaller but appeared anatomically normal with normal glans and meatus at the tip. Patient said he was urinating only through the left sided larger penis. Median raphe of scrotum was displaced to right side. Abdominal ultrasonography was normal. There were no gastrointestinal or genitourinary anomalies. There was only one urinary bladder. Anal canal was patent without any fistulae connection.

Patient was planned for elective surgery. Both urethral orifices were catheterized easily and ended up in a single bladder. Cystoscopy was done though left side phallus and right side urethra was seen which was opening in bladder [Fig 2]. The opening was identified by feeding tube in right side phallus. Both ureteric orifices and trigone appeared normal [Fig 3]. Right sided phallus was excised. It was extending to bladder. The wound in scrotum was closed in layers [Fig 4]. Excised phallus was found to have two well developed corpora cavernosa and corpora spongiosum [Fig 5]. Post surgery the patient had no urinary complaints and no erectile dysfunction till 6 months follow up.

Fig 1: Patient with double phallus

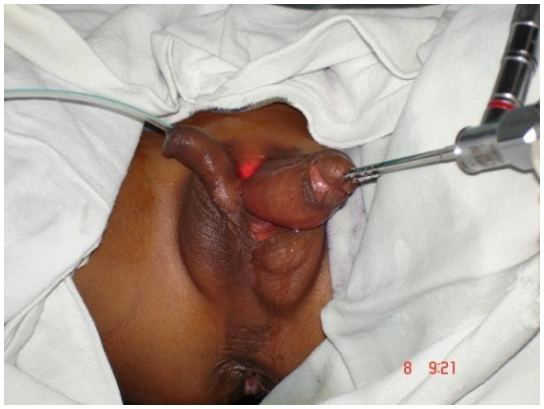

Fig 2: Cystoscopy to identify right urethral orifice

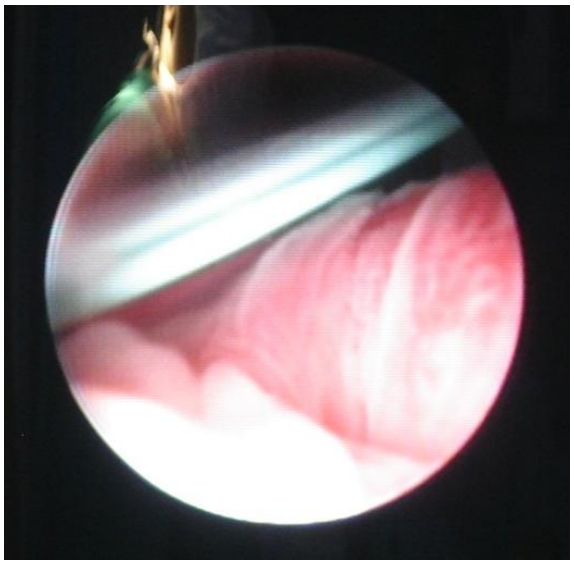

Fig 3: Cystoscopy view of accessory phallus

Fig 4: Wound in scrotum after excision

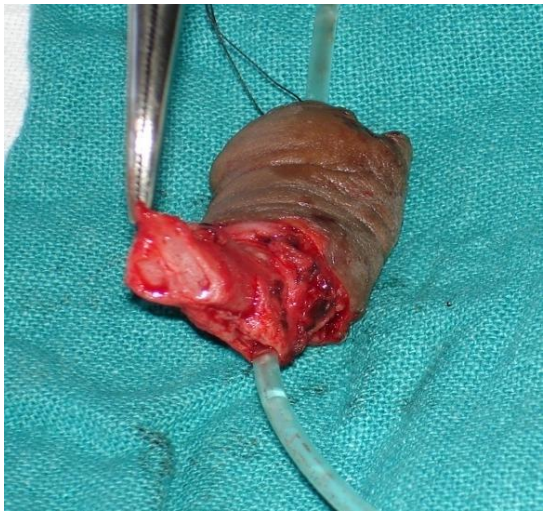

Fig 5. Excised phallus showing two corpora

DISCUSSION

Embryologically diphallus is believed to arise due to abnormal development of pubic tubercle. It could be due to either separation of pubic tubercles or cleavage of pubic tubercle. In former case each phallus will have only one corporal body and urethra but in later each phallus will have two corporal cavernosus bodies and urethras. [6] Caudal duplication syndrome has been proposed to explain the concomitant duplication of hindgut, bladder and urethra [6].

Schneider classified diphallus in three groups; diphallus of glans alone, bifid diphallus and complete diphallus. [7] Vilanova and Raventos have added a fourth category pseudodiphallus. [8] The urethra shows a range of variations from functioning double urethra to complete absence of urethra in each penis. A later classification currently widely accepted includes two main groups: true diphallia and bifid phallus [9]. These groups are further divided into partial and complete duplication. True complete diphallus means each phallus has two corpora cavernosa and corpora spongiosum. When this phallus is rudimentary or small it is called true partial diphallia. When only one corpora cavernosa is present in each phallus it is called bifid phallus. When degree of separation is to base of shaft it is called complete bifid phallus whereas if it is upto just glans, it is called partial bifid phallus. Our case was complete true diphallus.

True diphallia is more often associated with severe malformations as compared to bifid phallus. Associated anomalies include genitourinary anomalies which can be hypospadias, bifid scrotum, duplication of bladder and urethra, renal agenesis, extrophy alone or extrophy with vesicointestinal fistula[10]. Gastrointestinal anomalies can be imperforate anus with or without rectourinary fistula or duplication of colon. Musculoskeletal anomalies can be diastasis of pubis, club foot, polydactyly or lumbosacral anomalies [4].

Penile duplication poses a difficult treatment problem in terms of medical, ethical and aesthetic decision making. Through investigations are mandatory to reveal concomitant congenital malformations that is potentionally life threatening and require immediate surgical corrections. The treatment of diphallia is by excision of the duplicated noncommunicating penis. The treatment principally depends on the type of accompanying congenital abnormalities as well as preserving continence and erectile function which means individualizing each case. Surgical correction is individualized with the aims of achieving proper urinary continence, urinary stream and erection with adequate cosmesis.

Although isolated diphallus has been described in literature [4, 5, 6]. But in these cases penis had either hypospadiac meatus, pseudophallus or hypoplastic urethra [4, 6]. Our case has been unique that there was complete diphallia. Both phallus have two each corpora cavernosa and well developed corpora spongiosum. Although one phallus was smaller than the other, urethra was patent in both of them. Complete excision without erectile dysfunction and urinary dysfunction was possible.

CONCLUSIONS

Although isolated true diphallia can occur, it is very important to rule out other genitourinary anomalies which are common in these cases.

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