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DIPHALLIA – A COUNFOUNDING ANOMALY

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Abstract:

Diphallia or penile duplication is an extremely rare congenital anomaly. Its incidence is very rare, about one in every 5.5 million live births. The spectrum of presentation can vary from double glans only to complete duplication of penis with multiple anamolies. We report 4 years old boy with bifid glans small sized phallus with penoscortal hypospadias.

Key Words: Diphallia, penoscortal Hypospadias.

Introduction:

Diphallia or penile duplication is an extremely rare congenital anomaly occurring once in every 5.5 million live births (1-4) . In 1609, Wecker reported the first case of diphallia in Bologna, Italy(4). While only about 100 cases are reported in the literature till date(4-6). We report a case of 4 years old boy presented to us with bifid small sized phallus with penoscrotal hypospadias.

Case Presentation:

4 years old boy with normal birth history, accompanied by parents presented to outpatient department with complaint of abnormal meatal opening since birth. He had no past medical history. There was no urinary complaints. Parents had non consanguineous marriage. There is no significant family history of hypospadias or other congenital abnormalities. On examination there was duplicated glans and small phallus with penoscrotal hypospadias with severe chordee and hooded foreskin. Both testicles were normal and palpable in normal looking scortum. Ultrasound of the whole abdomen done did not reveal any developmental abnormalities. The patient initially underwent stage 1 hypospadias repair; where the chordee was corrected, and where the urethral plate was fashioned from the preputial foreskin. Since, the length of the penis was not adequate, 250 mg intramusucular testosterone injection was given once to the patient, six months later he had an adequate length of penile shaft and glans. The second stage of hypospadias was then undertaken; where, a v shaped excision was done to remove the excess ‘wings’ of glans. The uretheral plate had a u shaped incision on the shaft, to create the neo-urethra. Standard

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urethroplasty reinforced by a layer of subdartos fascia, glanuloplasty and meatoplasty, were then performed- giving the penis a postoperative look as shown in figure 2.

Discussion:

Diphallia is a rare anomaly. Very few cases are reported in literature and in each case presentation varies from a small accessory penis or duplication of the glans to complete penile duplication. The duplication may be orthotopic or ectopic. Division of the penis may be in sagittal or frontal plane. The shape and size can be symmetrical or asymmetrical (1, 3, 6) . The range of variation is seen in urethra as well, from complete absence of the urethra in each penis to functioning double urethra's(2, 6). In majority of cases there is a single corpus cavernosum in each organ. The meatus may be normal or can be associated with abnormality like hypospadias, or epispadias. The scrotum may be normal or bifid. The testicular abnormality includes athropic, or undescended testes(2, 4). Diphallia is divided into three groups by Scheider(2-4): Group I includes diphallia of the glans alone, Group II: bifid diphallia, and Group III includes complete diphallia. While fourth category named as pseudodiphallia later added by Villanova and Raventos(3). A newer classification which is currently widely accepted includes two main groups: true diphallia and bifid phallus (3, 4, 6) . Further subdivison of these two groups are partial or complete duplication. True complete diphallia is defined as complete penile duplication, each with two corpora cavernosa and a corpus spongiosum. While the term bifid phallus applies when each penis has only one corpus cavernosum. This type is further subdivided into complete bifid phallus when glans is complete separated till distal shaft of penis or partially involving the glans only, the anomaly is considered as partial bifid phallus. The term "pseudodiphallia," as originally discovered by Villanova and Raventos corresponds to true, partial diphallia (3) .

Diphallia is usually associated with other malformations, such as bladder extrophy. cloacal extrophy, anorectal malformation, colon and rectosigmoid duplication, abnormality of skeletal and heart muscles, ventral hernia, vertebral anomalies and pubic symphysis diastasis(1-4, 6-16). Child born with true diphallia are more prone to infection which leads to death as they are often associated with severe malformations compared with bifid phallus(3, 6). There is significant variation in degree of erectile function. Usually, one or both penises are capable of erection. In cases of true, complete diphallia simultaneous erection and occasionally, ejaculation have been reported when presented in advanced age. In contrast, normal function of the rudimentary phallus is rarely seen in case of pseudodiphallia(3, 6, 11).

In literature, the question how diphallia occurs is yet to be answered. Various possible embryological events are described by authors that result in diphallia but most are in consensus that this esoteric anomaly is may be due to defective fusion of the genital tubercle. While karyotype in majority of cases is normal(6, 11).

The treatment of diphallia requires surgical excision of noncommunicating duplicated penis(5, 6, 14, 17, 18) . While in case of complete true diphallia, the penile reconstruction needs to be done by combining corpora of each penis(2, 6, 16, 18).

Our case is complete bifid glans. The phallus was smaller in size and associated abnormality was penoscrotal hypospadias only. In literature majority of cases are found with true complete diphallia with various anomalies. Mirshemirani A-R *et al*. reported the case series of six cases with diphallia and out of six only one case has similar presentation to our case except he had bifid scrotum as well. He was managed surgically with phalloplasty and hypospadias repair along with scrotoplasty(8).

Conclusion:

Diphallia is rare anomaly with diversity in presentation and associated anomalies. Each case requires careful evaluation and individualization before going for any surgical intervention. The aim of treatment should focus the correction of diphallia and accompanying congenital abnormalities as well as the preservation of cosmesis, urinary continence, stream and erectile function (1, 3, 6).

The authors declaration

The authors declare that there is no conflict of interests regarding the publication of this paper.

Conflicts of Interest

The authors whose names are listed immediately above certify that they have NO affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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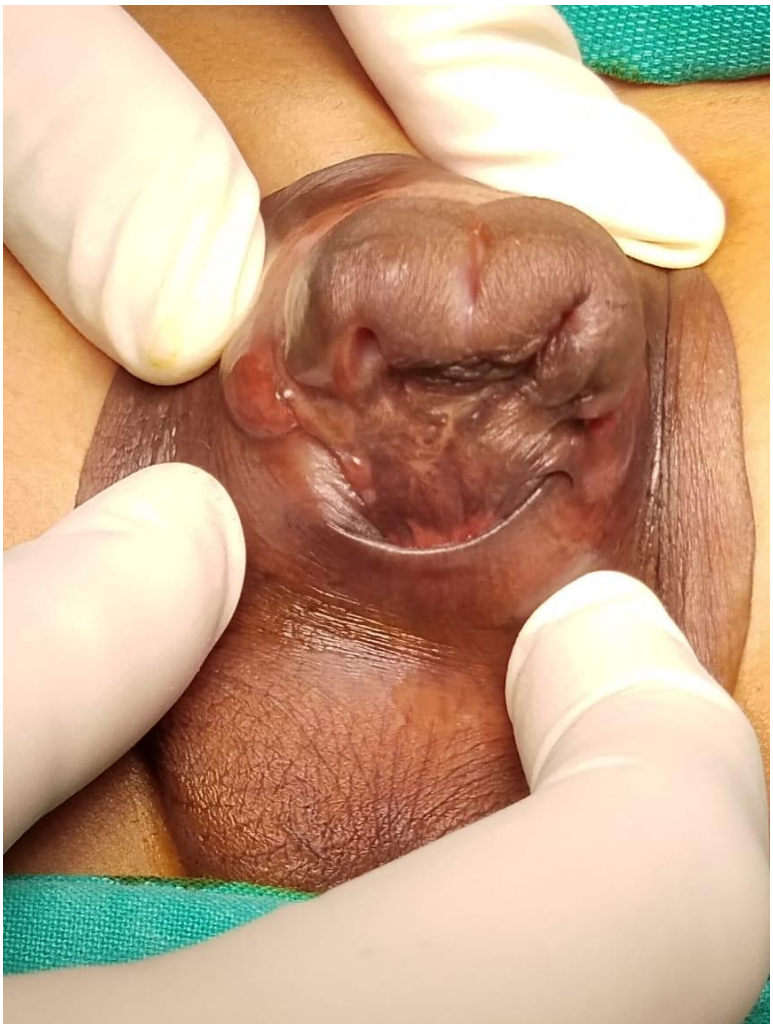

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