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Hypospadias associated with partial bifid phallus: A case report

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ABSTRACT

Diphallia is an extremely rare congenital anomaly. Bifid phallus is a type of diphallia and is rarely recorded in published studies. According to the degree of separation of the penises at the base of the shaft or just at the glans, bifid phallus is further classified into complete or partial forms. Bifid phallus is often associated with hypospadias or part of the exstrophy–epispadias complex. We are really lucky to have successfully treated a 2-year-old patient with penoscrotal hypospadias combined with partial bifid phallus in the shaft. After the surgery, the patient had no issues with penile function.

1. Introduction

Diphallia (duplication of the penis) is an uncommon anomaly with an estimated incidence of 1 per 5 million live births.[1,2] In which, bifid phallus - a rare type of diphallia, is scarcely reported. The estimated number of published cases to date is about 100.[3] It often occurs with multiple anomalies,[1] such as ectopic scrotum, bifid scrotum, hypospadias, imperforate anus, bladder exstrophy, colon duplication, double bladder, and vertebral deformities.[1,3] We present a 2-year-old boy with hypospadias combined with an isolated bifid phallus who underwent successfully surgical repair.

2. Case report

A 16-month-old boy was referred to our department for preoperative evaluation of penoscrotal hypospadias. His antenatal and family histories were unremarkable. Physical examination revealed a urethral meatus at the base of the ventral penis, the penis length was 1.9 cm, the glans diameter was 1.1 cm, bilateral testes were presented in the respective scrotal sacs. The foreskin is still attached to the glans on the dorsal side leading to bifid phallus was not recognized. After separating the foreskin, we found that the glans split to the collum glandis penis, and the penis was divided in the coronal plane into two moieties at the urethral orifice (Fig. 1). The patient has been diagnosed with a 46, XY karyotype. Abdominopelvic ultrasound and pelvic X-ray revealed no abnormality.

We performed surgical correction when he turned 2 years old. After penile degloving, no chordee was found by artificial erection, the posterior corpus cavernosa and spongiosum had a normal appearance, a partial bifid phallus in the shaft. Dorsal attachment of both cavernosa was performed by 6–0 absorbable suture The urethra is reconstructed by the urethral plate distal to the urethral orifice by 7–0 absorbable. A dorsal subcutaneous flap from the prepuce was used protective intermediate layer. We used a urinary catheter with balloon and removed it after 9 post-operative days. The penis achieved a good aesthetic shape after surgery (Fig. 2). The boy was discharged after 10 days with uneventful post-operative course.

Long-term follow-up: The boy was followed up 9 months post-operatively with 3 follow-up visits at 1 month, 6 months and 9 months. The child has good cosmetic results, good urinary stream and continence (Fig. 3). As noted by his parents, his penis has an erection in the morning.

3. Discussion

The first case of diphallia was reported in 1609 by J J. Wecker.[3] Following which approximately 100 cases have been reported in the literature with varied presentations for over 400 years.[1,2] Many classifications have been proposed for diphallia. In 1972, A classification which was proposed by Aleem[3] is being widely accepted currently, includes 2 main types: true diphallia and bifid phallus. These 2 types can be subdivided into 2 subclasses: complete and partial duplication. In

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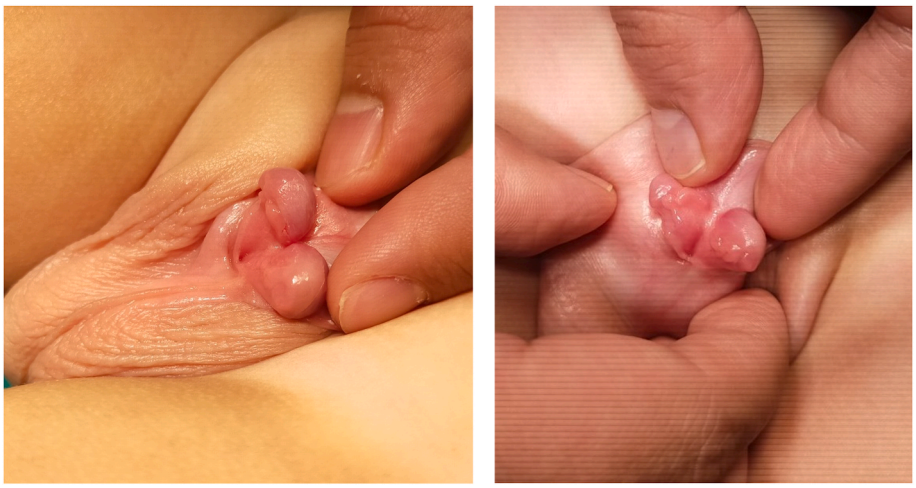

Fig. 1. Clinical appearance of bifid penis (Pre-operative).

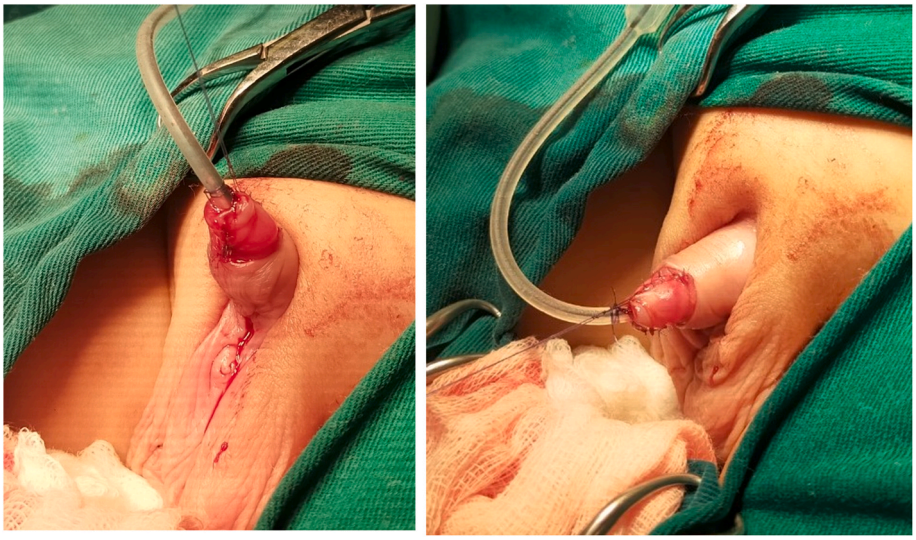

Fig. 2. Post-operative aspect of the reconstructed penis.

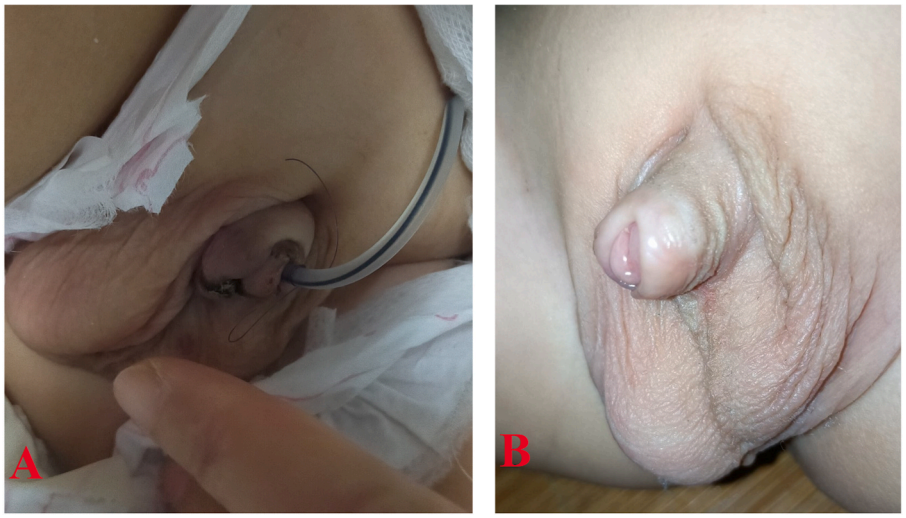

Fig. 3. 3A) 7th post-operative day with urinary catheter; 3B) The shape of the penis 6 months after surgery.

2017, Jessus[4] proposed a new simplified classification based on the clinical and surgical implications of each type: True penile duplication (each duplicate penis has 2 corpora and 1 spongiosum); hemiphalluses (each penis has corpora and a hemiglans); pseudo-duplication (normal penis with an accessory penis-like tissue); and partial duplication (duplication involves only the distal penis). The type presented in our case belongs to the partial bifid phallus and the group of hemiphalluses. according to Jessus's classification.[4]

The embryological causes for diphallia have not been fully understood. The normal development of the phallus begins with the coalescence of bilateral cloacal tubercles at the anterior end of the pars phallic of the urogenital sinus. Columns of mesoderm growing rapidly around the lateral margins of the cloacal plate form the genital tubercle.[1] If this hypothesis is followed, bifid phallus should be the more common form. However, in current literature,[1,2,5] reports of true diphallia are dominant in published studies, while little is known about bifid phallus or isolated cases. True diphallia is often associated with more severe malformations compared to bifid phallus. On the other hand, bifid phallus is often associated with hypospadias or epispadias,[5] or more severe malformation, such as exstrophy bladder. In the review of diphallia by Gyftopoulos[1] with 77 cases recorded, there were 50 cases of true diphallia and 27 cases of bifid phallus.

In cases of bifid phallus, according to the degree of separation of the penises at the base of the shaft or just at the glans, bifid phallus is further classified into complete or partial forms, respectively.[1] Matsumoto's[5] case involved a complete bifid phallus with a hypospadiac meatus in the perineum and pubic diastasis. After surgery, despite the good aesthetic record, the patient had urinary incontinence. Our patient did not have urinary incontinence. Thus, the complete form should have a worse prognosis than the partial form.

All the authors[4,5] who have published articles about diphallia have suggested that management of diphallia is challenging. In general, combination defects are usually processed first. Therefore, apart from careful examination should suggest some diagnostic testing such as: chromosomes, ultrasonography, voiding cystourethrography, magnetic resonance imaging, and computed tomography can be used to assess the associated internal anomalies, such as bladder and urethra duplication, kidney anomalies, and colorectal duplication. We agree with the classification of Jessus[4] which is easier to understand and more suitable for choosing the proper operative techniques for diphallia. Following the instructions, we choose to directly resect the hypoplastic duplicate penis, glans, or the accessory penile-like tissue in order to keep the main urethra. For hemiphalluses, many authors suggest joining the 2 penises without excision. In our case, after ruling that no chordee by artificial erection. The urethra is reconstructed by the urethral plate distal to the urethral orifice, in combination with hypospadias repair and epispadias repair. A dorsal subcutaneous flap was used to reduce urethral fistula complications. Unlike Matsumoto,[5] who performed the preputial free graft technique. Surgical correction is individualized to achieve proper urinary continence, urinary stream, and erection with adequate cosmesis.

4. Conclusion

Partial bifid phallus is an extremely rare congenital anomaly and often associated with hypospadias. Treatment method is grafting the 2 penises together and urethral reconstruction to achieve good aesthetic results and good penile function.

Patient consent

The child's legal gaurdian's consent was obtained to publish this case report.

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Ethical approval

The study was approved by our research committee, Viet Duc University Hospital, Hanoi, Vietnam.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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