



## مدیریت جراحی دیفالی در کودک ۱۰ ساله دارای نقص مادرزادی: یک گزارش موردی

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چکیده	اطلاعات مقاله
<p><b>زمینه:</b> دیفالیای یکی از نادرترین ناهنجاری‌های مادرزادی در مردان است که به وجود آلت تناسلی دوگانه منجر می‌شود. با وجود این که تداوی معمولاً شامل مداخلات جراحی است، هر مورد دارای پیچیدگی‌های خاص خود است. این مقاله به ارائه یک گزارش موردی از دیفالیای در یک پسر ۱۰ ساله و مدیریت جراحی آن می‌پردازد.</p> <p><b>معرفی موضوع:</b> مریض با وجود دو آلت تناسلی بستری شد. بررسی‌های یورولوژیکی نشان داد که مجرای ادراری سمت راست کامل و عملکردی است، در حالی که مجرای سمت چپ ناقص بود. مریض تحت جراحی قرار گرفت. جراحی شامل جداسازی مجاری و ایجاد یک مجرای واحد، همراه با برداشتن آلت سمت چپ به دلیل عملکرد ناقص و نگرانی‌های وجاهت آن بود.</p> <p><b>نتیجه گیری:</b> پس از جراحی، عملکرد ادراری مریض به‌طور کامل بهبود یافت و نتایج وجاهت قابل قبول بود. این مطالعه بر اهمیت تشخیص زودهنگام، ارزیابی دقیق و مداخلات جراحی مناسب برای مدیریت ناهنجاری‌های ولادی می‌پردازد و نشان می‌دهد که پس از جراحی، عملکرد ادراری مریض به‌طور کامل بهبود یافت و نتایج زیبایی‌شناختی قابل قبول بود.</p>	<p><b>نوع مقاله:</b> گزارش موردی</p> <p>تاریخ دریافت: ۱۴۰۳/۱۲/۱۰</p> <p>تاریخ پذیرش: ۱۴۰۴/۰۱/۲۸</p> <p>تاریخ نشر: ۱۴۰۴/۰۱/۳۰</p> <p><b>*شناخت‌نامه نویسنده مسؤول:</b></p> <p>احسان الله رسولی.</p> <p>دیپارتمنت جراحی اطفال، شفاخانه تخصصی آریا آپلو، هرات، افغانستان.</p> <p>✉ Ehsanrasouli6@gmail.com</p> <p><b>کد اختصاصی مقاله / DOI:</b></p> <p><a href="https://doi.org/10.58342/ghalibMj.V.2.I.1.13">https://doi.org/10.58342/ghalibMj.V.2.I.1.13</a></p>

**واژه‌گان کلیدی:** دیفالیای، تشوش ولادی، جراحی اصلاحی.

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## Surgical Management of Diphallia in a 10-Year-Old Child with a Congenital Anomaly: A Case Report

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Article Information	Abstract
<p><b>Type:</b> Case Report</p> <p>Received: 28/ 02/ 2025 Accepted: 17/ 04/ 2025 Published: 19/ 04/ 2025</p> <p><b>*Present address and corresponding author:</b> Ehsanullah Rasouli.</p> <p>Department of Pediatric Surgery, Aria Apollo Specialty Hospital, Herat, Afghanistan ✉ Ehsanrasouli6@gmail.com</p> <p><b>DOI:</b> <a href="https://doi.org/10.58342/ghalibMj.V.2.I.1.13">https://doi.org/10.58342/ghalibMj.V.2.I.1.13</a></p>	<p><b>Background/introduction:</b> Diphallia is one of the rarest congenital anomalies in males, resulting in duplication of the external genitalia. Although treatment typically involves surgical intervention, each case presents unique complexities. This article provides a detailed case report of diphallia in a 10-year-old boy and its surgical management.</p> <p><b>Case presentation:</b> The patient presented with two glans penises under the prepuce. Urological investigations revealed a functional complete right-sided urethra, while the left urethra was incomplete. Surgical correction included separating the urethras and creating a single functional channel, along with amputation of the left glans due to its incomplete functionality and aesthetic concerns.</p> <p><b>Conclusion:</b> Postoperative outcomes demonstrated full recovery of urinary function and acceptable aesthetic results. This case underscores the importance of early diagnosis, precise evaluation, and appropriate surgical interventions in managing congenital anomalies.</p>

**Key words:** Diphallia, Congenital Anomaly, Corrective Surgery

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## Introduction

Diphallia or also called diphallus is a malformation of the male urogenital system characterized by a complete or partial duplication of the penis. It is a rare congenital anomaly with an incidence of 1 per 6-5 million live births. About a hundred cases have been reported in the medical literature. This malformation can be associated with a duplication of the urinary system (ureters, bladder, and urethra) or the lower part of the digestive tract (ileum, appendix, colon, anal imperforation). More complex malformations such as exstrophy of the bladder, hypospadias, cryptorchidism and atresia of the esophagus can also be associated with diphallia<sup>[1]</sup>. It is a result of embryonic development of urogenital tract from paired ducts<sup>[2]</sup>.

## Treatment

Diphallia is treated surgically, but in each case, it is necessary to take into account the concomitant pathology. The main cause of death in children with diphallia are concomitant severe developmental defects, so the treatment is carried out in stages<sup>[3]</sup>.

Surgery is the only treatment option for diphallia. Treatment is not always necessary, however.

A surgeon will usually perform this surgery at birth or very soon after. The procedure will vary based on how much duplication there is and the presence of other birth irregularities.

Because each case of diphallia is unique, the surgery to treat it can be complicated and difficult. The primary concerns are:

- making sure that the male will be able to urinate normally and have erections
- reducing the potential risk of infection
- reducing structural irregularities

The timing of the surgery will be an important factor due to the male's likely age. As doctors often diagnose diphallia at birth, there may need to be several surgeries over time.

Individuals with diphallia often have other birth irregularities, such as hypospadias, duplicated urethras, and cryptorchidism (wherein one or both testes do not descend). Researchers report that in many cases, surgeons can repair the physical irregularities associated with diphallia<sup>[4]</sup>.

Penile duplication, or diphallia, is a rare congenital anomaly with a frequency of 1 in 5,000,000 neonates in the United States. The exact pathophysiological cause of this phenomenon is unclear; however, it seems that some teratogenic factors may interfere with the normal embryological development of the genitourinary system between the 23rd and 25th days of gestation.

During the embryology period, columns of mesoderm at the end of the urogenital sinus around the lateral aspect of the cloacal palate grow to form the genital tubercle. Failure in the fusion of the bands of these mesoderm causes bladder exstrophy and epispadias. Consequently, for embryological occurrence of penile duplication, longitudinal duplication of the cloacal membrane should occur to form the 3 or 4 columns of primitive mesoderm around the 2 cloacal membranes to finally form the two genital tubercles.

Although diphallia may appear as an isolated anomaly, it may also be associated with caudal duplication syndrome or as an exstrophy-epispadias complex. It may also be associated with multiple congenital malformations such as ventral herniation, musculoskeletal anomalies, anorectal malformations, and heart abnormalities.

Based on new clinical classification by Lisieux E groups, four types of penile duplication can be observed with various outcomes:

- 1: True diphallia (described as 2 separated penises with 2 corpora and one spongiosum body in each one, which is usually associated with caudal duplication syndrome).
- 2: Hemiphalluses (with 2 halves of the penile shaft, each one containing the hemiglans and a corpus; this type is a part of exstrophy-epispadias complex)<sup>[5]</sup>.

## Classification

Schneider 80 has previously classified diphallia into four main categories:

- (1) Duplication of the glans alone
- (2) Bifid diphallia
- (3) Complete diphallia with each penis having two corpora cavernosa and a corpus spongiosum
- (4) Pseudodiphallia in which there is a rudimentary accessory atrophic penis existing independently of the normal penis<sup>[6]</sup>

## Case Report

A 10-year-old boy was admitted to the pediatric surgery ward for evaluation of duplicated penis, a condition present since birth. On physical examination, two glans penises were observed under the prepuce. Both glans had a distinct urethral orifice, and Foley catheters were easily passed through each orifice.

When asked to urinate, the patient voided primarily from the right-sided urethral orifice, with minimal urine passing from the left side. A detailed urological evaluation, including cystoscopy, revealed a complete urethra on the right side, while the left urethra was incomplete, connecting to the prostatic urethra on the right side.

## Surgical Procedure

The patient was scheduled for surgical correction. Intraoperatively, the left urethra was carefully separated from the right up to the level of the prostatic urethra. A side-to-side anastomosis of the urethras was performed to create a single functional urethral channel. Additionally, the left glans penis was amputated due to its incomplete functionality and aesthetic concerns.

## Discussion

Diphallia is a rare anomaly, and it is believed that no cases are identical. It varies from a small accessory penis or duplication of the glans to complete penile duplication. The duplication, each with two corpora cavernosa and a corpus spongiosum. When the duplicate penis is smaller or rudimentary, it corresponds to true partial diphallia. When only one corpus cavernosum is present in each penis, the term bifid phallus applies. When the degree of separation is complete to the base of the shaft or to just the glans, the anomaly is considered complete or partial bifid phallus, respectively. The term "pseudodiphallia," as originally described by Villanova and Raventos, corresponds to true, partial diphallia. Our case is a complete bifid phallus.

Diphallia or penile duplication is a rare anomaly, with an estimated prevalence of 1 in every 5.5 million live births. It may present alone or associated with other penile or abdominal malformations, such as renal and anorectal anomalies. At the genetic level, defects in the homeobox genes may be involved<sup>[7]</sup>.

Neugebauer in 1898 and Nesbit and Bromme in 1933 reviewed cases of diphallus in literature, after the first case reported by Weker in 1609.

Although only 100 cases were reported in literatures, we believe that this is merely an underestimation as many of these are unrecorded or not reported to the surgical practice<sup>[8]</sup>.

Another case with imperforate anus, double bladder, and duplicated penis, reported by Mirshemirani et al<sup>[9]</sup>.

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. True diphallia is more often associated with severe malformations compared with bifid phallus. Infants born with diphallia and its related conditions have higher death rate from various infections associated with their more complex renal or colorectal systems. In cases of bifid phallus and orthotopic, true, and complete diphallia, both

phalli are attached to the pubic bone; otherwise in the cases of pseudodiphallia or true, partial diphallia with an ectopic penis, the corpora are usually not attached to the pubis. In this case, amputation of the accessory penis is easier.

Following a literature review, each diphallia patient is a unique variant with its own anatomical configuration and associated anomalies<sup>[10]</sup>.

The degree of erectile function in cases of diphallia varies significantly. Usually, one or both penises are capable of erection. In cases of true, complete diphallia presented at advanced age, simultaneous erection and, occasionally, ejaculation have been reported. In contrast, pseudodiphallia is rarely associated with normal function of the rudimentary phallus. Several studies reported normal erection of partial bifid phallus and true partial diphallia even though erectile function after surgery is still unclear<sup>[11]</sup>.

## Conclusion

The patient tolerated the procedure well and had an uneventful recovery. Postoperative follow-up showed normal urinary function, with urine passing entirely through the right urethra. No complications were observed, and the patient was discharged with a good prognosis.

Postoperative outcomes demonstrated full recovery of urinary function and acceptable aesthetic results. This case underscores the importance of early diagnosis, precise evaluation, and appropriate surgical interventions in managing congenital anomalies.

## Conflict of Interest

We express our profound gratitude to the dedicated teams from the Pediatric Surgery and Urology Surgery departments for their valuable contributions to this work. Additionally, we extend our sincere appreciation to Ghalib Medical Journal for their support and for publishing this case report.

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