Picture story

A case of diphallia with anorectal malformation

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Introduction

Diphallia is an extremely rare urological anomaly with a reported incidence of 1 in 5–6 million live births[1]. It can present as complete or incomplete diphallia, varying from a small accessory penis or duplication of the glans to complete penile duplication[2]. In most cases it is associated with complex urological, gastrointestinal, or anorectal malformations[3]. Newborns with diphallia and related conditions face an elevated risk of mortality due to various infections linked to the intricacies of their renal or colorectal systems[4]. The treatment of diphallia is by excision of the duplicated non-communicating penis[5]. We report a newborn who presented on the 3rd day of life with diphallia and anterior ectopic anus.

Case report

A baby boy was born to non-consanguineous parents at term. Mother was a 20-year-old who was in her second pregnancy without any complications such as gestational diabetes mellitus or pregnancy induced hypertension. She has not had pre-conceptional folic acid and was not anaemic. Her ultrasound scans, including anomaly scan, did not reveal any significant fetal anomalies. The baby was delivered by normal vaginal delivery at 39 weeks of gestation with an Apgar score of 10 at 1st, 5^{th,} and 10th minutes. The baby weighed 3.2kg (median) with length of 46cm (-1SD to -2SD) and occipito-frontal circumference of 36cm (median). Baby had passed urine and meconium within the first 24 hours of life.

Phallus 1 was 2.5 cm in length and was situated on the right side of the perineum while phallus 2 was 1.5 cm in length and was situated on the left side of the perineum (Figure 1). Both phalluses had normal-shaped glans with 2 urethral openings located un the normal position. However, he was passing urine from the right phallus only. He had a single scrotum and well-formed rouge with palpable testes.

Ultrasonography of the urological system showed normal kidney and bladder morphology. Both penises had normal internal morphology, but a normal urethra was seen within the large right penis. Both were attached to the posterior surface of the pubic bone. The micturition cystourethrogram showed a normally functioning right urethra which was connected to the bladder base in an eccentric right-sided location. He had an anterior ectopic anus with normal tone without faecal incontinence, which was located at the base of the scrotum (Figure 2).

Figure 1: Phallus 1 and phallus 2

Figure 2: Anterior ectopic anus shown by arrow

His 2D-echocardiogram and spinal x-rays were normal. He was examined under anaesthesia on day 5 of life and it was planned to remove the extra non-functional phallus with correction of anterior placed anus with anorectoplasty at 4 months of age as there is a better response to operative therapy when anoplasty is performed prior to 6 months of age.

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Discussion

Diphallia is a rare congenital anomaly comprising two structurally and anatomically separate phalluses. A widely accepted classification separates true diphallia from bifid diphallia. It can be further divided into complete or partial duplication. In true diphallia each phallus has two corpora cavernosa and a corpora spongiosum. Bifid phallus has only one corpora cavernosum in each penis[6]. Diphallia is commonly associated with other congenital anomalies such as hypospadias, ectopic scrotum, duplication of bladder, imperforate anus and vertebral anomalies[6]. As diphallia presents with a spectrum of diseases, extensive investigations are required to identify associated congenital malformations. Treatment should always be individualized according to the degree of penile duplication and extent of associated anomalies[4]. Management of these patients is a challenge to the multidisciplinary team[4].

References

1. Deshpande V.. Complete diphallia - Our technique to avoid complications. *Journal of Indian Association of Pediatric Surgeons* 2020; **25**(3): 182-3. https://doi.org/10.4103/jiaps.JIAPS_76_19 PMid: 32581449 PMCid: PMC7302456
2. Zhang W, Yu N, Liu Z, Wang X. Pseudo-diphallia: a rare kind of diphallia. *Medicine* 2020; **99**(33): e21638. https://doi.org/10.1097/MD.0000000000021638 PMid: 32872026 PMCid: PMC7437754
3. Agzamkhodjayev S, Ergashev K, Abdullayev Z, Soliyev A, Batrutdinov R. Complete duplication of the penis - A case report. *Urology Case Reports* 2022; **40**: 101892. https://doi.org/10.1016/j.eucr.2021.101892 PMid: 34745893 PMCid: PMC8551595
4. Habib M, Bajwa HF, Abbas M, Chaudhary MA. A very rare case of diphallia with anorectal malformation. *International Journal of Surgery Case Reports* 2023; 105: 107980. https://doi.org/10.1016/j.ijscr.2023.107980 PMid: 36933406 PMCid: PMC10031018
5. Aparicio-Rodríguez JM, Cuellar-López F, Hurtado-Hernández ML, *et al.* Disorders of sexual development in genetic paediatrics: three different ambiguous genitalia cases report from hospital Para el Nino Poblano, Mexico. *International Journal of Genetics and Molecular Biology* 2010; **2**(10): 207–16.
6. Tirtayasa PMW, Prasetyo RB, Rodjani A. (2013). Diphallia with associated anomalies: a case report and literature review. *Case Reports in Urology* 2013; 2013: 192960. https://doi.org/10.1155/2013/192960 PMid: 24383036 PMCid: PMC3870645
7. Hassan MHSM, Basnayake S. (). A rare case of Diphallia associated with anorectal and vertebral malformations. *Sri Lanka Journal of Perinatal Medicine*, 2020; **1**(1), 40. https://doi.org/10.4038/sljpm.v1i1.29