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Ventral partial diphallia associated with hypospadias: A case report

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ABSTRACT

Diphallia, or penile duplication, is a rare congenital urological malformation. It may be associated with other congenital malformations. The objective of present paper is to report the case of a two-year boy with ventral duplication of the penis associated with proximal hypospadias. The ventral penis was amputated, and Duckett urethroplasty was done. There were no reported postoperative complications in the follow-up.

1. Introduction

Diphallia, also referred to as penile duplication, is a rare congenital anomaly of mal external genital organ. It's estimated to occur in 1 out of 5–6 million births. Since first publication of Jacob Wecker in 1609, around 100 patients being reported within the literature.[1] Clinical presentation of cases of diphallia differ greatly from one another, varying from double glans with a common shaft, to complete duplication of the phallus.[2] It often occurs with multiple anomalies, such as ectopic scrotum, bifid scrotum, hypospadias, imperforate anus, bladder exstrophy, colon duplication, double bladder, and vertebral deformities.[3] We report this case and it particularity followed in our hospital and review of the literature concerning the management and follow-up of this condition.

2. Case report

A 2-year-old male child was received in our hospital with abnormal penis. He was born to a 30-year-old mother, gravida 4 and para 3. In the past history of his mother, we didn't found use of any teratogen drugs. At birth, the patient had no vital distress. Physical examination revelated the presence of two partially separate penises of different sizes, one in front of the other. The ventral penis was smaller. We noted one urethral meatus at the base of the ventral penis, bilateral testes were presented in the respective scrotal sacs (Fig. 1). The foreskin is still attached to the dorsal glans on the dorsal side. No permanent urine flow occurred from meatus. Abdominal ultrasound showed normal renal and urinary tract. Retrograde urethrocystography revealed a single bladder with one urethra meatus without vesicoureteral reflux (Fig. 2). Uroscan was normal. Ventral pseudodiphallia with proximal hypospadias was diagnosed.

We performed surgical correction at 2 years old and 3 months. Accessory penis excision was performed and sent for anatomopathological examination. Its dimensions were 2cm × 0.8cm × 0.5cm. After penile degloving and excision of chordee, artificial erection was normal. One catheter was placed through the urethra and across the anastomosis. The urethra was reconstructed according to Duckett's technique with an entire pedicled tube, by 7-0 absorbable suture, and penis ski coverage according to Byars with the rest of foreskin (Fig. 3). Urethral catheter was removed on the 10th and he was discharged the same day. After a 4-year follow-up, the child has good cosmetic results and was fully continent with normal voiding. Histological examination of accessory penis reveals a corpus cavernosum.

3. Discussion

Diphallia is a rare condition, with an estimated incidence of 1:5 million live births.[1] Although the anomaly is known from the XVII century, there are only just over 100 cases reported so far.[4] Many classifications have been proposed for diphallia. Schneider[5] classification is

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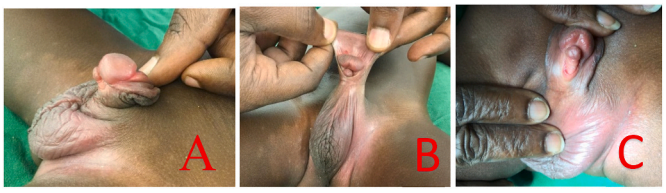

Fig. 1. Diphallia with two partially separate penises of different sizes, one in front of the other. A: Profile view. B: Front view with foreskin. C: Front view without foreskin.

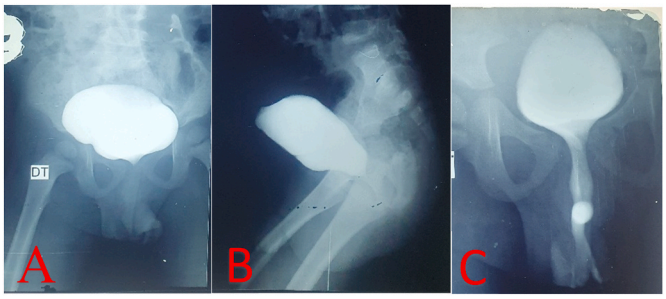

Fig. 2. Normal cystography. A: frontal cystography. B: profile cystography. C: permictional imaging.

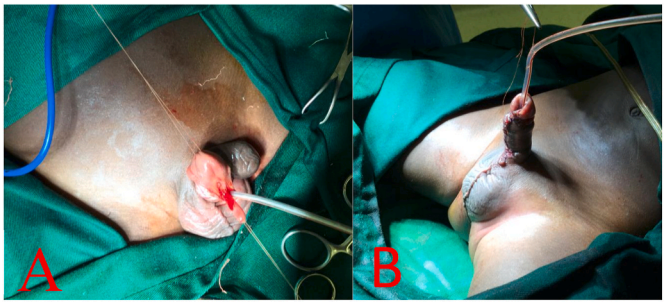

Fig. 3. D: Intraoperative picture. E: Immediate postoperative appearance.

the one that is generally used and has classified diphallia into three main categories: Duplication of the glans alone; Bifid diphallia; Complete diphallia with each penis having two corpora cavernosa and a corpus spongiosum; Vilanova and Raventos[6] have added a 4th category namely Pseudodiphalia in which there is a rudimentary accessory atrophic penis existing independently of the normal penis. According to modified Schneider's classification our patient would be considered pseudodiphallia but ventral. Histological examination of accessory penis concluded a corpus cavernosum only. Its particularity was its anterior location and the association with hypospadias. Cases of diphallia differ greatly from one another. Other anomalies may coexist, such as the ectopic scrotum, hypospadias, double bladder, renal and gastrointestinal malformations, imperforate anus, colon duplication, and vertebral deformities.[2] The malformative assessment include generally: x-ray for pubic and lumbosacral exploration, Abdominal and urinary tract ultrasound, voiding cystourethrogram, and M.R.I. which gives more information about the corpora cavernosa and corpus spongiosum and their path. Also it allows complete anatomical study of this malformation. In our case, no cases of associated malformations were found. Urethrocystoscopy must be realized just before surgical intervention and can provide some information about urethra, bladder and ureteral meatuses.

4. Conclusion

Dphallia is a rare urological condition which can be associated in very rare condition with hypospadias. Its management isn't easy and requires careful study of the type of diphallia to determine adequate surgical approach. Whatever the anatomical type of diphallia, a surgical approach must to ensure a good cosmetic results, normal erectile function and fully continent with normal voiding.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Declaration of competing interest

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Abbreviations

M.R.I.: Magnetic Resonance Imaging.

Authorship

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

CRediT authorship contribution statement

Kouame Soroboua Agbara: Conceptualization, Investigation, Methodology, Writing – original draft, Writing – review & editing. **Adjoba Manuela Ehua:** Data curation, Formal analysis. **Helen Audrey Thomas:** Data curation, Investigation. **Reine Christine Akproh:** Data curation, Visualization. **Jean-Marie Konan:** Formal analysis, Visualization. **Yapo Guy Serge Kouame:** Visualization. **Ouattara:** Investigation. **Ahou Sekinatou Oyewola:** Visualization. **Martial Olivier Moulot:** Formal analysis, Supervision, Visualization. **Roumanatou Sanni Bankole:** Supervision, Validation.

References

1. Kendrick DJ, Kimble RM. Diphallia: literature review and proposed surgical classification system. *ANZ J Surg*. 2022;92:2053–2065.
2. Duarte ML, dos Santos LR, da Silva AQP, da Silva MQP. Diphallia: a case report of a rare anomaly evaluated by magnetic resonance imaging. *Turk J Pediatr*. 2021;63(5): 917–921. https://doi.org/10.24953/turkjped.2021.05.021.
3. Frolov A, Tan Y, Rana MW-U-Z, Martin JR. A rare CASE of human diphallia associated with hypospadias. *Case Rep Urol*. 2018;2018. https://doi.org/10.1155/2018/8293036.
4. Schneider P, Lamitter JK, Uson AC, Melicow AC. The male genital tract. In: Mustard WT, Ravitch MM, Snyder WH, et al., eds. *Pediatric Surgery*. second ed. Chicago: Year Book Medical Publishers; 1969:1263.
5. 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.
6. Vilanova X, RaventoS A. Pseudodiphallia, a rare anomaly. *J Urol*. 1954;71:338–346.