



Case Report

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Diphallia, a rare congenital anomaly- A case and literature review

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Abstract

Diphallia or also called diphallus is a malformation of the male urogenital system characterized by a complete or partial duplication of the penis. The authors report one rare case of a newborn baby transferred from pediatrics for better management of a polymalformative syndrome associating diphallia and anal imperforation are treating in Urology unit at general surgery department, Centre hospitalier universitaire departemental Borgou-Alibori, at Parakou City.

Keywords: Diphallia, Congenital anomaly, Parakou.

INTRODUCTION

Diphallia or also called diphallus is a malformation of the male urogenital system characterized by a complete or partial duplication of the penis [1]. It is a rare congenital anomaly with an incidence of 1 per 5-6 million live births [1, 2]. About a hundred cases have been reported in the medical literature [3]. 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 [2].

This pathology leads to a modification of the body pattern and consequences on reproduction in the event of delayed treatment, but also a significant psychological impact and a risk of social exclusion, especially in our African context.

The rarity of cases of diphallia in Africa but also the interest in sharing the experience of sequential management of cases of diphallia associated with other malformations in under-equipped settings motivates this work.

Observation

This is a newborn baby transferred from pediatrics for better management of a polymalformative syndrome associating diphallia and anal imperforation.

The gestational history shows a pregnancy well followed with five prenatal consultations during which 3 ultrasounds were performed and no morphological abnormalities were found.

A malaria during pregnancy was noted during the 16 weeks of amenorrhea managed by an artemisinin-based combination. A serological test (toxoplasmosis, rubella, hepatitis) was requested again but not honored by the patient.

He was born at term following a eutocic delivery. The urogenital examination finds the presence of two well-individualized penises, fused at their base with a single scrotum (Image 1A). After scaling, the penises are located side by side, the supernumerary being to the right of the principal. The external urethral meatus is in an apical position (Image 1B). The urethral catheterization of the main (left) penis is easy and brings back clear urine; that of the right penis abuts at the peno-scrotal level (one-eyed). In addition, both testicles are present in the bursae and an anal imperforation is observed.

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Image 1A: Diphallia before cracking



Image 1B: Diphallia after cracking

Abdominopelvic ultrasound and abdominopelvic CT scan (with and without injection) found two kidneys in normal position, two ureters and a single bladder. Uretrocystoscopy could not be performed due to the unavailability of a pediatric endoscopy column. The colostogram showed an intermediate-type malformation with a normal-looking sacrum. Serum creatinine, uremia, and cytobacteriological examination of the urine were normal.

He received an emergency colostomy and then admitted 6 months later for the management of diphallia which consisted of excision of the supernumerary penis (Images 2A to 2B).

Patient under general anesthesia, a vertical incision passing through the median groove of the ventral surface of the supernumerary penis up to the peno-scrotal junction is made, removing the supernumerary penis to the base allowing to individualize its urethra and its two bodies cavernosa, ligature then section of the supernumerary penis at its base for reimplantation to the main penis.

Suture the subcutaneous tissue with Vicryl 3/0 at separate stitches, then the subject skin with Vicryl 2.0.



Image 2A: Dissection of the supernumerary penis



Image 2B: Excision Limit of the penis

Macroscopically, the surgical specimen showed a penis about 3 cm long with the presence of two corpora cavernosa, a gland and a urethra ending in an apical meatus.

A Charriere 6 Foley catheter is left in place for 48 h. The postoperative follow-up is simple and the infant is released 4 days later after the first dressing has been made.

Reviewed two weeks after discharge, healing is good and urination is satisfactory and a morning erection is maintained. Finally, an anoproctoplasty was scheduled three months later.



Image 3A: Supernumerary penis



Image 3B: Aspect at the end of the operation



Image 4A: Supernumerary penis



Image 4B: Aspect at the end of the operation

DISCUSSION

With an incidence of 1 case per 5 to 6 million live births, diphallia is a rare urogenital malformation [4]. About a hundred cases have been reported worldwide, particularly in Europe [1, 5], America [6, 7] and Asia [8, 9]. Several theories attempt to explain the embryology of diphallia [10]. It would either result from a "separation" of the pubic tubercles, in which each phallus has corpora cavernosa and a urethra, or from a "cleavage" of the pubic tubercle in which each phallus has 2 corpora cavernosa and a urethra [11]. The three main factors implicated are exposure to endocrine disrupting drugs, infections or homeopathic genes [12]

In our case, only artemisinin was taken during pregnancy and a theoretical link has not been established between this drug and the occurrence of fetal malformations. However, no serological tests (HIV, VDRL-TPHA, Rubella, HBc, HBs) have not been performed during pregnancy, much less the search for homeopathic genes. The absence of these examinations limits the search for an etiological investigation.

Although extremely rare, diphallia can also affect the female sex and consists of a duplication of the clitoris [13].

There are several classifications on diphallia including that of Schneider which includes three types: diphallia of the glans, bifid diphallia and complete diphallia [1]. Vilanova described a 4th type called pseudodiphallia [14]. The French Association of Urology (AFU) has proposed three types of diphallia according to their external morphological and histological appearance: complete and incomplete true diphallia, complete and partial bifid penis and pseudodiphallia. [1]. A new classification recently proposed by Lisieux et al [15] was based on embryological, anatomical, clinical and therapeutic aspects to classify diphallia into true diphallia, hemidiphallia, pseudodiphallia, and partial duplication. Taking into account the classification of AFU, the infant in our study had incomplete true diphallia.

Diphallia can be associated with other malformations such as bladder duplication, urethral duplication, bladder exstrophy, colonic duplication, scrotal bifidity [16]. True diphallia is most often associated with severe abnormalities compared to the bifid phallus [16, 17]. In fact, anal imperforation is more frequent in cases of complete diphallia [10, 13], which is corroborated by our clinical case. Ectopic urethral discharge such as hypospadias or epispadias may be present with penile duplication [18]. The urethral meatus of both penises were in an apical position. In the series by Mirshemirani et al. [10] in six cases of diphallia (including five true diphallias), hypospadias was present in two, epispadias in one, and three had an apical urethral meatus. The bifidity of the scrotum reported in some studies [10, 19] was not found in our case.

Treatment of diphallia involves excision of the supernumerary penis and surgical correction of associated abnormalities [10, 20]. The more complex the associated malformations, the more delicate the management will be. In any case, the treatment must meet 3 requirements, which are: the preservation of continence, the preservation of erectile function and the achievement of a satisfactory aesthetic result [16, 21, and 22].

Management of associated abnormalities can be done during the neonatal period and excision of the supernumerary penis a few months later or even at infancy [5]. This therapeutic approach was the one reported by Mirshemirani et al. [23] for a case of true diphallia associated with a duplication of the bladder and an anal imperforation which required at third day of life, a colostomy then at the age of 4 months, the excision of the supernumerary penis and of the bladder with cystoplasty and left ureterovesical reimplantation. In our case, the anal imperforation required an emergency colostomy on the same day at third day of life and then 6 months later, we performed the excision of the supernumerary penis. In Africa, pathologies of the genital tract are considered "taboo" according to religious and socio-cultural beliefs. Excision of the supernumerary penis during childhood is a possible option, but at risk given the social and psychological repercussions that this presents. Indeed, our child's diphallia can be a reason to laugh at the part of other children in school during recess.

CONCLUSION

Diphallia is a rare urogenital anomaly whose diagnostic approach requires careful clinical examination and exhaustive imaging work to identify associated abnormalities. This is the 1st case of diphallia in Benin. Surgical treatment consisted of an emergency colostomy and excision of the spare penis 6 months later. Surgical treatment must meet a threefold imperative: the preservation of continence, the preservation of the erection and the achievement of an aesthetic penis.

Conflicts of interest

The authors declare no conflict of interest, the article being read and approved by all.

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