

Anterior Sagittal Epispadias Urethral Duplicity: A Case Report

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Abstract

Case Report

Urethral duplication is a rare congenital urological anomaly that can be detected incidentally or due to urinary symptoms in young patients. Symptoms may include recurrent urinary tract infections, dysuria, bifurcated urinary stream, urinary incontinence, and purulent discharge from the accessory meatus. The pathophysiology remains poorly understood, with various embryological theories proposed. Treatment typically involves surgical excision of the accessory urethra, especially in symptomatic cases. Urethral duplication is often associated with other malformations, and Effman's classification system categorizes it into three types based on the location of the accessory urethra. We present the case of a 4 years old boy with anterior sagittal epispadias urethral duplicity.

Keywords: Urethra, duplicity, children.

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INTRODUCTION

Urethral duplication is a rare congenital condition that predominantly affects boys, with only a few reported cases in girls [1, 2]. There have been numerous attempts at classification, including those by Stephens, Cendron, Williams, Effman, and Das, despite which it remains poorly defined, as there may be confusion due to differing terminology [3]. Urethral duplicity can manifest as either complete or incomplete, and may or may not be accompanied by other urogenital anomalies such as hypospadias, epispadias, or bladder exstrophy [4].

CASE REPORT

We report the clinical observation of a 4-year-old patient who is consulting for a urethral duplicity which was first noticed following circumcision performed at the age of 2 years.

- The patient had no history of recurrent urinary tract infection or dysuria.
- Genital examination revealed a circumcised penis.

The patient had two urethral openings: one had a normal size and positioned orthotopically at the glans,

while the other, smaller in size, was located dorsally on the penis (epispadias), approximately 1 cm proximal to the normal urethra. A scrotal examination was normal.

The patient underwent a radiological exploration such as UCG which showed a bladder of normal size and morphology, without wall abnormalities, with no vesico-ureteral reflux and no double urethral system.

Also, the patient underwent cysto-urethroscopy which led to the diagnosis of an anterior sagittal epispadias urethral duplicity: it showed a normal urethra, intact external urethral sphincter, and normal prostatic urethra. The bladder neck and bladder were also visualized and found to be normal.

We concluded to an anterior sagittal epispadias urethral duplicity

The Surgical Procedure:

The cannulation of the accessory urethra showed a blind ending accessory urethra. Two catheters were inserted through both meatus: the normal and the ectopic (fig 1).



Figure 1: the cannulation of both the meats: the normal and the ectopic

Next, we made a longitudinal incision from the ectopic meatus to the path of the Foley catheter. The supernumerary urethra was dissected back to its origin

and then resected (fig2),(fig 3). The closure was done with vicryl 5/0 wire.

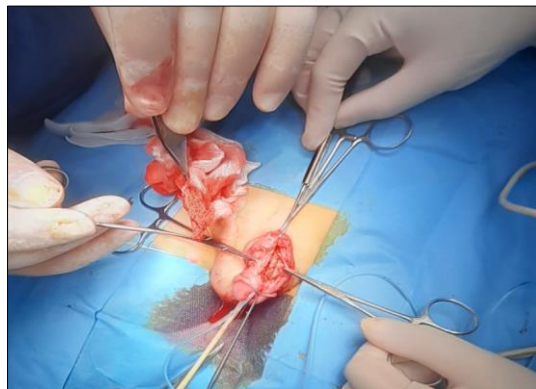


Figure 2: Dissection of the ectopic urethra

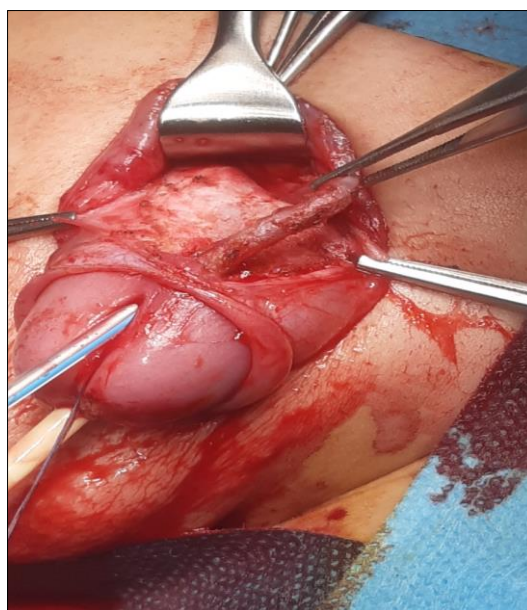


Figure3: Dissection of the urethra to its blind end

DISCUSSION

Urethral duplication is a rare congenital urological anomaly. Its detection may occur incidentally in asymptomatic cases or may be prompted by nonspecific urinary symptoms in typically young patients. Literature reports various symptoms associated with urethral duplication, including recurrent urinary tract infections, dysuria, bifurcated urinary stream, urinary incontinence, and purulent discharge from the accessory meatus [5, 6].

The pathophysiology of urethral duplication (UD) remains poorly understood. Various theories have been proposed to explain the different anatomoclinical types, but none have provided conclusive evidence. However, authors generally concur on treating only symptomatic cases of UD. Surgical excision of the accessory urethra, possibly in conjunction with correction of associated malformations, has shown to yield improved outcomes [5].

Urethral duplication (UD) is frequently associated with other malformations [6], which should be carefully assessed. These may include penile curvature, epispadias, hypospadias, phimosis, cryptorchidism, buried penis in the scrotum, penoscrotal transposition, bladder exstrophy, Prune Belly Syndrome, and persistent peritoneal-vaginal canal. Various theories based on embryology have been proposed to explain UD, but none have emerged as predominant [8, 9]. According to Das and Brosman [10], the embryological anomaly in organogenesis involves the opening of the urogenital sinus at the urogenital diaphragm. An extension thereof, known as the urogenital blade, faces the genital tubercle and is absorbed to form the urogenital groove. The prostatic urethra and membranous urethra originate from this blade. Occlusion of the groove, from posterior to

anterior, leads to the formation of the penile urethra, externally marked by a median raphe. This represents a failure in regression of the Müllerian ducts, culminating on the dorsal aspect of the urogenital sinus. Meanwhile, Woodhouse and Williams [9], and Merrot T [11], suggest an ischemic process during...

In a literature review, we identified four different classification systems for this anomaly. The classification system by Effman *et al.*, describes three types of urethral duplication based on the radiological location of the accessory urethra (fig 4).

In Type I, there is a blind-ending accessory urethra (incomplete urethral duplication), which is further classified into two subclasses: Type I-A and I-B. In Type I-A, the distal duplicated urethras open on the dorsal or ventral surface of the penis but do not communicate with the urethra or bladder. In Type I-B, a rare condition, a proximal-accessory urethra opening originating in the urethral channel ends blindly in the peri-urethral tissues.

In Type II, there is a completely patent accessory urethra, classified into two subtypes: A when there are two meatuses and B when there is one. Type II A is further classified into two subtypes: IIA1 and IIA2. In IIA1, two noncommunicating urethras arise independently from the bladder, while in IIA2, a second channel arises from the first and courses independently into a second meatus in a Y-shaped manner (Y-type). In Type II-B, two urethras arise from the bladder or posterior urethra and unite into a common channel distally.

In Type III, an accessory urethra arises from a duplicate or septate bladder [7].

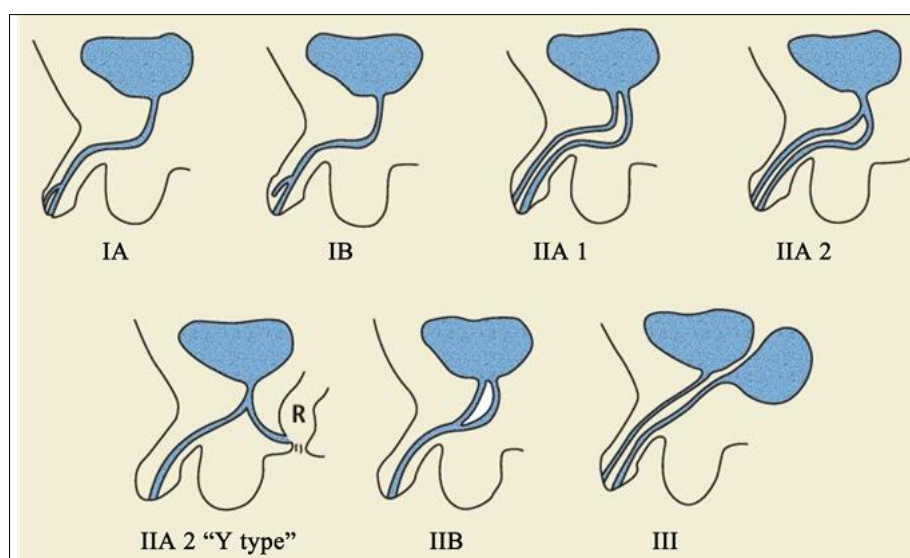


Figure 4: Effman classification system of UD

When symptoms of urethral duplication (UD) are pronounced, authors concur that surgical intervention is necessary, involving total resection of the accessory urethra [11, 12].

Most authors recommend simple resection of the accessory urethra, without including epispadias specific curing technics [11, 12].

CONCLUSION

Urethral duplication is a rare congenital malformation typically seen in young adult males and may coincide with other genital anomalies. Despite ongoing research, the exact causes and mechanisms of urethral duplication remain unclear. Efforts to refine the classification system, such as Effman's, are ongoing, with the need for additional subtypes. Surgical intervention is the primary treatment for symptomatic urethral duplication, involving the removal of the accessory urethra.

Conflict of Interests: The authors have no conflict of interests to declare.

REFERENCES

1. Arena, S., Arena, C., Scuderi, M. G., Sanges, G., Arena, F., & Di Benedetto, V. (2007). Urethral duplication in males: our experience in ten cases. *Pediatric surgery international*, 23, 789-794.
2. Wani, S. A., Munianjana, N. B., Jadhav, V., Ramesh, S., Gowrishankar, B. C., & Deepak, J. (2019). Urethral duplication in children: experience of twenty cases. *Journal of Indian Association of Pediatric Surgeons*, 24(4), 275-280.
3. Tolrá, J. R., Campaña, J. M. C., Buisan, O., Comalat, A. F. I., Miranda, E. F., & Milá, N. S. (2005). Duplicidad uretral incompleta. *Actas Urológicas Españolas*, 29(7), 697-699.
4. Rimtebaye, K., Sillong, F. D., Tashkand, A. Z. A., Kaboro, M., Niang, L., & Gueye, S. M. (2015). Complete Urethral Duplicity: A Rare Cause of Urinary Incontinence, New Type According to Effman's Classification. *Open Journal of Urology*, 5(10), 182.
5. Rimtebaye, K., Sillong, F. D., Tashkand, A. Z. A., Kaboro, M., Niang, L., & Gueye, S. M. (2015). Complete Urethral Duplicity: A Rare Cause of Urinary Incontinence, New Type According to Effman's Classification. *Open Journal of Urology*, 5(10), 182.
6. Ben Nsir, R., Jouini, R., Maazoun, K., Helal, Y., Krichéne, I., Mekki, M., & Nouri, A. (2008). Epispadiac urethral duplication in boys. *Progrès en urologie*, 18(9), 570-574.
7. Suoub, M., Saleem, M. M., & Sawaqed, F. (2020). Complete urethral duplication: case report and literature review. *Research and Reports in Urology*, 15-20.
8. Slavov, C., Donkov, I., & Popov, E. (2007). Case of duplication of the urethra in an adult male, presenting with symptoms of bladder outlet obstruction. *European urology*, 52(4), 1249-1251.
9. Woodhouse, C. R. J., & Williams, D. I. (1979). Duplications of the lower urinary tract in children. *British journal of urology*, 51(6), 481-487.
10. Das, S., & Brosman, S. A. (1977). Duplication of the male urethra. *The Journal of urology*, 117(4), 452-454.
11. Merrot, T., Weber, D., Steyeart, H., & Allessandrini, P. (2002) Sagittal Duplication of the Urethra in Children. Report of 7 Cases (Duplications sagittales de l'urètre chez l'enfant. À propos de sept observations). *Progrès en Urologie*, 12, 77-83.
12. Jouini, R., Krichéne, I., Lefi, M., Chelly, S., Mekki, M., Belghith, M., & Nouri, A. (2003, October). Urethral duplication in boys: about 4 cases. In *Annals of Urology* (Vol. 37, No. 5, pp. 288-292). Elsevier Masson.