

Duplication of Hypospad of the Urethra Associated with Bulbar Stricture in a Young Man aged 29 Years: Case Report from the Urology Unit of the Bamako Commune I Reference Health Center

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DOI: [10.36347/sjmc.2024.v12i03.002](https://doi.org/10.36347/sjmc.2024.v12i03.002)

| Received: 15.01.2023 | Accepted: 27.02.2024 | Published: 03.03.2024

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Abstract

Case Report

Urethral duplicity is a rare congenital malformation most commonly found in boys. Many anatomical forms have been described. The form associated with urethral stricture is a fairly rare combination. We report a case of hypospad urethral duplicity associated with bulbar urethral stricture. The patient was a 29-year-old male who presented with a double stream of urine associated with dysuria. On completion of the clinical and paraclinical examinations, we made the diagnosis of Effmann and Lebowitz type IB hypospade urethral duplicity associated with narrowing of the bulbar urethra. We proceeded to section the mucosal partition separating the 2 urethras from the meatus up to the bifurcation zone, followed by endoscopic internal uretrotomy of the stricture. Follow-up was straightforward and the result was deemed satisfactory after two years.

Keywords: Urethra, duplication, stricture, malformation.

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INTRODUCTION

Duplicity of the urethra is an extremely rare congenital malformation [1]. It may extend from the neck of the bladder to the tip of the glans and may be complete or incomplete. It is more common in boys and occurs preferentially in the sagittal plane [2]. Numerous anatomical varieties have been described, with the second urethra opening in a normal or non-normal position. Several classifications have been proposed, but the one described by Effmann and Lebowitz is currently one of the most widely used (Figure 1) [3].

The clinical expression of urethral duplicity is variable, ranging from a simple double urine stream to

recurrent urinary tract infection, dysuria or even complete retention of bladder urine. Retrograde micturition urethrocytography (RMUC) is the complementary examination of choice, providing a morphological study of the main urethra and possibly the supernumerary urethra [4].

Management is often difficult and depends on the anatomical type. The form associated with urethral stricture is an even rarer combination. To our knowledge, very few similar cases have been reported in the past. We report the case of a 29-year-old man with duplication of the urethra associated with bulbar stricture.

Citation: Berthe, A, Ballo, B, Drago, A. A, Kone, O, Sissoko, I, Ouattara, A. D, Maiga, A, Haidara, K, Diarra, I, Coulibaly, M. T. Duplication of Hypospad of the Urethra Associated with Bulbar Stricture in a Young Man aged 29 Years: Case Report from the Urology Unit of the Bamako Commune I Reference Health Center. Sch J Med Case Rep, 2024 Mar 12(3): 243-246.

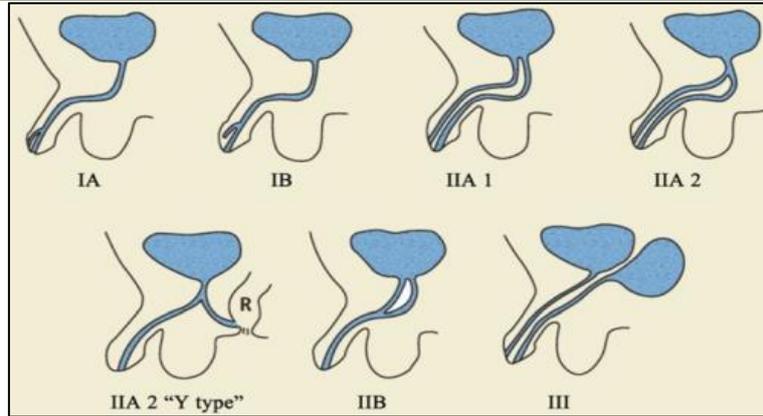


Figure 1: Classification of Effmann and Leibowitz [3]



Figure 2: View during consultation Arrows indicate the urethral meatus

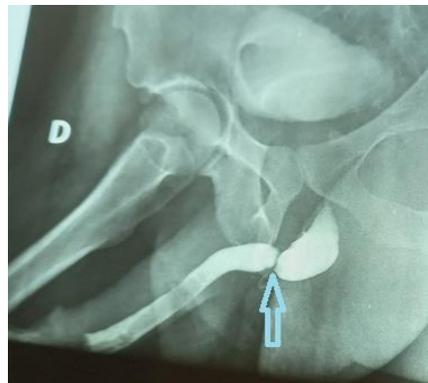


Figure 3: UCRM scan showing narrowing of the bulbar urethra



Figure 4: Arrows indicate urethral hypospad postoperatively



Figure 5: View at three months

OBSERVATION

Mr. O.B, 29-year-old male admitted to the department for double urinary stream, dysuria. In the history, it was noted: recurrent urinary infections, no notion of trauma or curvature of the penis upon erection. The history of the disease was marked by dysuria associated with a double and weak urination stream with sometimes delayed drops. On clinical examination, the patient was in good general condition; on the glans two (2) orifices were noted, one of which was in the hypospadias position. Figure 2. Preoperative view: arrows indicate the 2 meatus. Simultaneous catheterization of the two meatus identified the site of the malformation.

During urination, the urinary flow from the hypospadias meatus was weak but drip urination could be seen at the apical meatus. The remainder of the examination was normal.

Various additional examinations were carried out in particular: - Renovesico-prostatic ultrasound with evaluation of post-micturition residue which revealed moderate bilateral uretero-hydronephrosis

The Cytobacteriological Examination of Urine (ECBU) with antibiogram had isolated *Escherichia coli* treated with ofloxacin 200mg. The rest of the biological examinations were normal. After proof of the sterility of the urine, a retrograde and voiding urethrocytography (UCRM) was done without the images being of quality for an unequivocal interpretation. The urethrocytography could not be completely carried out due to the narrowing of the urethra but made it possible to know the level of the bifurcation located between the balano-preputial fold and body of the penis. At the end of this examination, we retained the diagnosis of hypospadias urethral duplication type IB according to Effmann and Lebowitz associated with a stricture of the bulbar urethra. Figure 3 and 4. No other malformation was associated.

After obtaining the patient's free and informed consent, the surgical procedure was performed under

spinal anesthesia. It consisted of a section of the mucous membrane separating the two urethras, from the urethral meatus to the bifurcation zone, then an endoscopic internal urethrotomy of the stricture. A charriere 20 silicone urinary catheter was then placed for 15 days. The postoperative course was simple and the patient left the center after 4 days. Two weeks after removal of the probe, a bimonthly calibration protocol was established for 3 months then monthly for 6 months. At the end of these 9 months of calibration, the urinary stream was judged satisfactory. After two years of follow-up, no complaints were reported, urination remained normal and the cosmetic appearance of the penis was judged to be good.

DISCUSSION

Urethral duplication is an extremely rare congenital malformation. It is more common in boys [1, 2]. In 2008, fewer than 500 cases had been reported in the literature [5, 6]. Many theories have certainly been put forward to explain the mechanisms of occurrence of DU, but there are still gray areas due to the multiplicity of anatomical varieties [7, 8]. Anatomically, the DU can be sagittal, collateral or even posterior. It can be epispadias or hypospadias [6,8]. The case that we reported in the present work is hypospadias DU associated with narrowing of the bulbar urethra.

Several classifications have been proposed but those of Williams-Kenawi and Effmann/Lebowitz are widely used [11]. The case reported here presented characteristics compatible with hypospadias urethral duplication type IB according to Effmann and Lebowitz. However, in this case there is a narrowing of the associated bulbar urethra. We did not find any other associated malformation. Some authors such as Faustin Mouafo Tambo *et al.*, [9], Pippi Salle JL *et al.*, [10], noted the existence of a complex association of malformations, with an incontinent anus, agenesis of the coccyx and disjunction of the symphysis. pubic. DU is a malformation whose diagnosis should be early but can be made at any age of life. However, the average age of

diagnosis is 29 months [2, 9, 10, 11]; our case was diagnosed at 29 years old.

This could be explained by the poor socio-economic conditions and illiteracy of the family. Urethral duplication is generally discovered incidentally during examination of the external genitalia of the newborn. Most cases are asymptomatic (7) however if it is symptomatic, the signs commonly encountered are the presence of a double urinary stream, a double urethral meatus, urinary incontinence, recurrent urinary infection, bending of the penis [1, 5, 7]. In our case it was dysuria associated with a double urinary stream and a double urethral meatus.

Diagnosis is based on clinical examination of the external genitalia. Diagnostic confirmation is based on the coupled UCRM - Uretrocystoscopy which will determine the anatomical type. Ultrasound looks for very often associated abnormalities and comorbidities such as dilation of the upper urinary tract [1, 12].

The management of urethral duplication is not yet well codified, the therapeutic attitude varies from one author to another. Only symptomatic forms require surgery [9-11]. Before any surgical procedure, it is imperative to clarify the anatomy and course of the two urethras.

Several therapeutic modalities have been used, including dilation of the normal urethra, injections of sclerosing substances into the supernumerary duct considered dangerous due to the risk of fibrosis of the corpora cavernosa according to certain authors [1, 9]; surgical excision of the accessory urethra, etc. In our case we first proceeded with a section of the mucous membrane separating the two urethras, from the urethral meatus to the bifurcation zone, then in a second step with an endoscopic internal urethrotomy of the urethral stricture. Sectioning of the mucous membrane separating the two urethras had also been done in cases reported by certain authors [5, 12]. According to I. Diabaté et al [8], this therapeutic modality exposes not only the risk of ejaculatory disorders, but also the persistence of the accessory urethra because it is not resected.

A favorable postoperative outcome is generally described, as was also observed in our case.

CONCLUSION

Hypospadiac urethral duplication is a rare congenital malformation of the urinary system whose main symptomatology is the presence of a double urinary stream. The diagnosis and precision of the anatomical type are essentially based on UCRM. Conventional surgical treatment and/or endoscopy generally guarantees a good long-term result.

Conflict of interest: The authors declare that they have no conflict of interest.

Sources of funding: None

Author contributions: All authors actively contributed to the conduct of this work, from care to the writing of the article. They all declare having read and approved the final version of the manuscript.

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