Acta Med. Okayama, 2003
Vol. 57, No. 2, pp. 91-93

Acta Medica Okayama
http://www.lib.okayama-u.ac.jp/www/acta/

Case Report

Urethral Duplication

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Urethral duplication is a rare congenital anomaly. Although a number of theories have been proposed to describe the embryology of the condition, the actual mechanism of the disorder is still unclear. We report here a case of urethral duplication in a 11-year-old boy complaining of a double stream, and review the current literature on this rare entity.

Key words: urethra, abnormalities, duplication

Urethral duplication is a rare congenital anomaly with approximately 188 cases described in literature [1]. This anomaly is most common in males with few cases reported in females.

Embryology of the condition is unclear. Several embryological theories have been proposed. Casselman and Williams [2] stated that a partial failure or an irregularity of the ingrowth of the lateral mesoderm between the ectodermal and endodermal layers of the cloacal membrane in the midline accounts for the forms with a dorsal epispadiac channel. Das and Brosman [3] reported that abnormal termination of the müllarian duct was responsible for urethral duplication. Rica *et al*. [4] suggested that asymmetry in the closure of the urorectal septum results in an urethraperineal fistula. In spite of the numerous theories proposed to explain this anomaly, no single theory explains all the various types of anomalies.

Received July 18, 2002; accepted November 12, 2002.
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Case Report

We report here a case of urethral duplication in a 11-year-old boy complaining of a double stream. He had no difficulty with micturition. Physical examination revealed a normal healthy boy with no other associated abnormality. There was a normal meatus at the apex of the glans and a secondary meatus about 10 mm proximally on the ventral aspect of the glans. A voiding cystourethrogram showed a double urethra as far as the membranous urethra (Fig. 1). Surgical operation was planned but the patient opted out of treatment, since he was asymptomatic and had no clinical problems except for a double urinary stream.

Discussion

There have been several attempts to classify urethral duplications. Gross and Moore [5] described this anomaly as a complete second passage from the bladder to the dorsum of the penis or as an accessory pathway that ends blindly on the dorsal or ventral surface. Effman *et al*. [6] reported classified urethral duplication into 3 types. Type I-Incomplete urethral duplication (A. Distal / B.

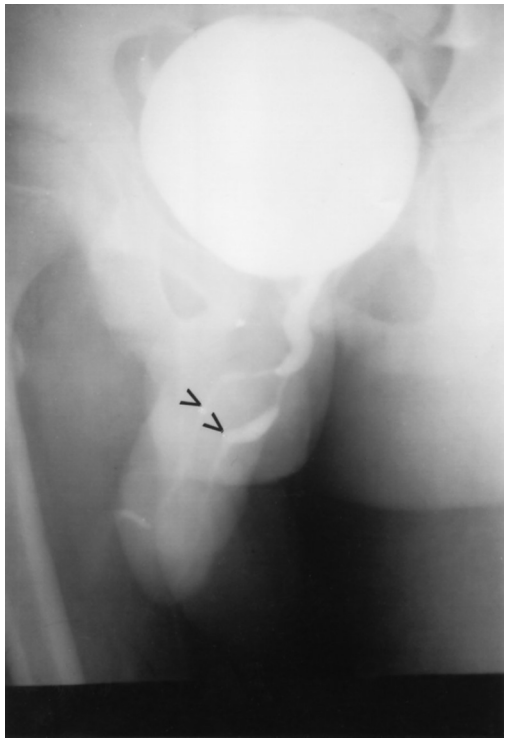

Fig. 1 Voiding cystourethrography shows complete urethral duplication.

Proximal), Type II-Complete urethral duplication (A. 2 meati / B. 1 meatus), Type III-Urethral duplication as a component of partial or complete caudal duplication. Das and Brosman [3] classified duplicated urethra into 3 types. Type I is a complete accessory urethra arising from a separate or confluent opening within the bladder and extending to an external orifice. Type II includes accessory urethras that arise from the primary urethra and may or may not extend to a distal orifice. Type III arises from the bladder neck or prostatic urethra and opens onto the perineum. The main urethra may be atretic. Firlit [7] classified duplication as a urethra that arises proximally from the bladder, bladder neck or duplicated bladder. Its distal course usually is dorsal to the main urethra. The complete form extends from the bladder to the glans.

Most duplications occur in the same sagittal plane, one on top of the other. Less commonly, openings of urethral duplication lie collaterally (side by side) in the frontal plane. The classification of Williams and Kenawi [8] includes epispadiac, hypospadiac, spindle and collateral types. According to Urakami *et al.* [9], collateral urethral duplication in the frontal plane has been reported in only 9 cases in the literature.

The classification of Effman *et al.* [6] is the most

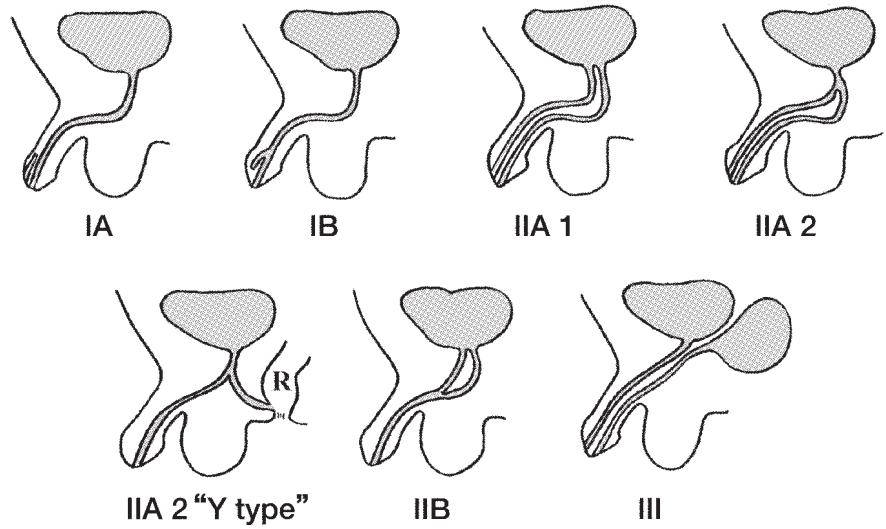

Fig. 2 Illustration of Effman classification. Type I, blind incomplete urethral duplication; Type IA (distal), opens on the distal or ventral surface of the penis but does not communicate with the urethra or bladder (most common type); Type IB (proximal), opens from urethral channel and ends blindly in the periurethral tissue (very rare). Type II, complete patent urethral duplication; Type IIA-1, two noncommunicating urethras arising independently from the bladder or coursing independently to 2 different meati; Type IIA-2, a second channel arising from the first and coursing independently to a second meatus; Type IIB (one meatus), two urethras arising from the bladder or posterior urethra and uniting to form a common distal channel. Type III, urethral duplication as a component of a partial or complete caudal duplication.

commonly accepted. This classification is the most functional, representing all clinical aspects of urethral duplication. According to the classification of Effman *et al.* [6], our case was a Type II A-2 (complete patent with 2 meatus; that is, a second channel arises from the first and courses independently to a second meatus).

The clinical significance of this abnormality is variable. Patients with complete urethral duplication can be asymptomatic or can present with a double stream, incontinence, recurrent infection or outflow obstruction. A double stream is the most common complaint and can be annoying, especially when the accessory urethra opens proximally on the penis, or it can be of little concern to the patient when the meatus are close together and the streams are parallel.

In conclusion, knowledge of urethral duplication is important with respect to surgical procedures. The treatment of urethral duplication depends on the anatomy of the duplication and its clinical manifestations. Some patients with mild symptoms do not require an operation. However, surgery should be considered for disturbing symptoms, such as an annoying double stream or incontinence, or for a cosmetic deformity, such as an epispadiac meatus.

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