Embryologic Preputial Sinus Remnant Mimicking Urethral Duplication

Salih Somuncu, Fatma Caglayan, Murat Cakmak and Sevgi Ulusoy

Department of Pediatric Surgery, Kirikkale University, School of Medicine, Kirikkale, Turkey

Abstract. Urethral duplications are rare congenital anomalies. Approximately 200 cases of urethral duplications have been reported in literature. We present a seven-year-old-boy with an embryologic preputial sinus remnant which was mimicking urethral duplication. The clinical presentations and embryology of the urethral duplication and preputial sinus are discussed. [Indian J Pediat 2006; 73 (3) : 227-228] E-mail : somuncusal@yahoo.com

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Duplication of the urethra is a rare congenital abnormality. It was first described by Aristotle.1 Approximately 200 cases of urethral duplications have been reported in literature.2 The most common form of this anomaly consists of a blind incomplete accessory urethra opening on the dorsum of penis. It is usually asymptomatic. Incomplete and complete urethral duplications frequently present with symptoms of incontinence, urinary infections, double stream but these are less common.2 The accessory urethra is rarely seen on the ventral surface of the penis.3 A case of embryologic preputial sinus remnant mimicking urethral duplication is presented.

CASE REPORT

A seven year old boy was found to have a penile defect during a routine physical examination. He had no complaints. A sinus 1 cm. in diameter and 2 cm. deep was opening on the ventral surface of the penis at subcoronal level. Urethral catheter was inserted into the sinus and it was found that the sinus was ending blindly in periurethral tissue and there was no communication with the urethra. (Figs. 1, 2) Other physical examination findings, routine urine, CBC and hormonal tests were normal. Abdominal ultrasonography and cystoscopic examination were normal. Intravenous urography revealed normal urinary system. Preputial sinus excision and circumcision was performed under general anesthesia. Recovery after surgery was uneventful. Histopathologically the sinus tract was reported to have squamous epithelium with slightly hyperkeratotic and
inflammatory vascular stroma. There were no urethral layer and uroepithelium. This histopathologic features revealed that sinus tract was the embryological remnant of prepuce.

DISCUSSION

Urethral duplications can be divided into three types according to Effman et al. Type 1A is most common and usually asymptomatic whereas Type 1B is very rare. Complete and incomplete urethral duplications (Type 2A and B) are less common but are clinically more significant. Symptoms include double stream, incontinence and urinary infection. Type 3 urethral duplications associated with other forms of partial or complete caudal duplication are very rare. The dorsal urethra is commonly the accessory urethra in cases of urethral duplication. The embryological explanation is the lack of mesodermal function in the formation of the genital tubercles or fusion that occurs too late or too far posteriorly. The accessory urethra rarely occurs ventral to the normal urethral channel. Singh and Murray presented a single case of distal urethral duplication. In the present case with preputial sinus ending blindly on the ventral surface of the penis which had no opening on the glans or in the periurethral tissue and didn’t contain urethral layers histopathologically. The embryology of the penile prepuce was described as progressively extending preputial fold which fused with epithelial covering of the glans and formed by a combination of preputial folding and ingrowth of a cellular lamella. This ingrowth creates the prepuce, glans, corona and coronal sulcus mucosa. The authors think that insufficiency of cellular lamella ingrowth may be the cause for an embryologic remnant such as preputial sinus like in the present case. Review of the available literature shows that this type of preputial sinus has not been reported earlier. It is recommended that this case should be considered in differential diagnosis of urethral duplications.

REFERENCES