



## Congenital fistula of the penile urethra: A case report

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**Abstract** Urethral fistulous opening to the penile shaft with or without chordee is extremely rare. Here, a 4-year-old boy with congenital urethral fistula is reported. English literature was reviewed for the former similar cases. Historical analysis showed that no appropriate nomenclature occurred for this isolated anomaly.

**Key Words** Child; penil urethra; urethral fistula; congenital urethral fistula.

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### INTRODUCTION

Congenital fistula of the penile urethra (CFPU) is extremely rare. As it may be an isolated fistula [1-7], several penile pathologies such as hypospadias and/or chordee may accompany [3,8,9]. Patients with CFPU are typically presented with “two way” micturition. Success of the surgical intervention depends on the additional deformities of the penis and the quality of the corpus cavernosum at the site of the fistula.

Here, a patient with congenital fistula of the penile urethra was presented by reviewing the literature.

### CASE REPORT

A 4 year-old boy with normally appearing meatus was presented with “two-way” urine flow during micturition. In physical examination, a fistulous opening at penile shaft with local defect in surrounding corpus spongiosum was detected. Glans penis and urinary meatus were normal. The patient was circumcised. No chordee was present (Fig. 1).

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*Accepted for publication 2 April 2014*

Urinalysis was found normal. Abdominal ultrasonography, intravenous pyelography and voiding cystourethrography revealed no pathologic findings. The patients prepared for surgery after routine laboratory tests. The fistula was repaired primarily by two layers fashion without urethral stenting. Postoperative course of the patient was uneventful. No recurrent fistula occurred. He was discharged at seventh day postoperatively. Urethral stricture requiring dilation was not detected at late postoperative period.



**Fig. 1.** Congenital fistula of the penile urethra, with intact prepucium and normal glans.

## DISCUSSION

The urethral plate is the anterior portion of the dorsal membrane of urogenital sinus. After ninth week of gestation external virilization occurs under the influence of testosterone and

dihydrotestosterone. At the end of 12<sup>th</sup> week the urethral plate folds inward to fuse in the midline, forming a tubularized urethra. The urethral plate is a lamellar cord of cells that represents the anterior surface of the urogenital sinus. Urethral folds develop or the mesoderm proliferates on either side of the urethral plate and unites to form the endodermal portion of the urethra, fusing from dorsal to ventral. This fusing represents formation of the urethra from a cranial to caudal direction up to the corona [10]. The glanular urethra is thought to be formed by an ingrowth of ectodermal cells from the tip of the glans in a caudal to cranial direction to meet the endodermal urethra, later canalizing to complete the urethral opening [11].

CFPU is an extremely rare condition and the etiology is controversial. Although the embryologic mechanism of the fistula still remains obscure, several theories have been proposed. Like many congenital defects, embryology may give clues about the occurrence of the CFPU. A CFPU may occur in the anterior urethra, where the spongiosum has been developed incompletely permitting a small diverticulum to form that can rupture antenatally [12]. Olbourne [7] speculated that a focal or temporary defect in urethral plate function

probably causes fistula located in the penile shaft.

The defect of corpus spongiosum in CFPU is similar with the defect that is encountered in hypospadias. Since the nonglanular hypospadias is caused by failure of urethral fold to unite over and cover the urethral groove [13], CFPU may be speculated as a variant of nonglanular hypospadias without glanular or prepuce defect [14]. The association of chordee and hypospadias in several CFPU cases may support this theory. According to Cook and Stephens [15], the formation of the fistula is resulted from pressure atrophy from the heel of the baby's foot, leading to pressure necrosis.

Wei et al. [16] have proposed another external compression theory. They reported a case of penile urethral fistula caused by a retained intrauterine device in 26 years-old man.

Various surgical techniques were described for CFPU. Pedicle flap [9], modified Denis-Browne urethroplasty [7], direct closure [7] and proximal based skin flap [5] techniques were defined. Maarafie and Azmy [4] converted the CFPU into a penile hypospadias and repaired by Byres skin flaps.

The primary complication of the repair is recurrent fistula formation [3,7]. The recurrent

fistula may heal spontaneously [7], or a subsequent operation to close the fistula may be required [3,7]. In the present case, we repaired CFPU primarily. Postoperative course of the patient was uneventful. No recurrent fistula occurred. These extremely rare cases should be carefully evaluated.

### CONFLICT OF INTEREST

**None declared.**

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