

# Congenital Prepubic Sinus with Remnant Tissue Mimics Corpus Spongiosum: A Rare Case

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## Abstract

### Keywords

- congenital prepubic sinus
- corpus spongiosum
- dorsal urethral duplication

A congenital prepubic sinus is a tract that originates in the skin and overlays the base of the penis; however, its embryologic basis is still debated. We herein present a case involving a 3-year-old boy who was admitted for examination of overlying tissue located a few centimeters distal to the dorsal base of the penis. Examination revealed a tiny sinus in the prepubic area, and 3 cm of tissue was attached to the sinus. Pathologic examination showed that the tissue was lined with squamous epithelium and continued along the sinus tract, which was lined with urothelial epithelium. According to Stephens' classification, the sinus appeared to be a variant of type 2 dorsal urethral duplication, and the remnant tissue mimicked the corpus spongiosum of the penis.

## Introduction

A small number of cases of congenital prepubic sinus (CPS) have been reported.<sup>1,2</sup> CPS is a rare congenital anomaly of the urinary tract. It is associated with discharge from an opening overlying the symphysis pubis, and affects both males and females.<sup>3</sup> CPS may either be a complete or incomplete channel that runs parallel to the normal urethra from the prepubic glans or clitoral area and extends to the bladder, which may either join the urethra or end blindly.<sup>4</sup> Dorsal urethral duplication and cloacal anomalies are assumed to be the causes of this condition.<sup>5</sup> In most cases of CPS, a tiny hole is seen in the prepubic area. We herein report a case of CPS in which the tissue extending from the overlying skin mimicked the corpus spongiosum of the penis.

## Case Report

A 3-year-old boy with weight of 12 kg was admitted for yellowish discharge from a tiny opening along the midline of the pubic–glans junction on the dorsal base of the penis. No

other symptoms related to the genitourinary tract were noted. All clinical and laboratory findings were normal. Physical examination revealed a tiny sinus in the prepubic area and 3 cm of tissue, similar to the corpus spongiosum, attached to the sinus. Ultrasound (US) examination showed a sinus directed toward the bladder neck. Fistulography showed a noncommunicating fistulous tract coursing from the prepubic skin toward the bladder (►Fig. 1).

During surgery, the overlying tissue located 3 cm from the base of the penis was excised and found to be continuous with the sinus tract. The sinus opening was probed and simple excision of the sinus tract was performed, revealing a tract approximately 4 cm long that extended to the retropubic space and ended in a fibrous tract. The postoperative course was uncomplicated, and the boy remains asymptomatic 1 year after the intervention. Histomorphological findings showed that the tissue was lined with squamous epithelium, and the accompanying urothelial residue was compatible with urothelial cysts and surrounded by focal smooth muscle layers. The entire sinus tract was lined with urothelial epithelium (►Fig. 2).

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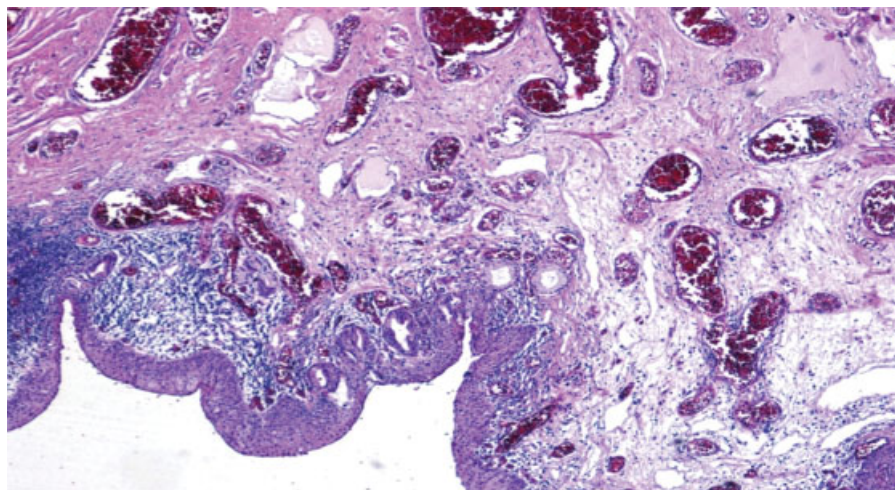
**Fig. 1** Prepubic sinus with tissue mimicking corpus spongiosum.

## Discussion

CPS is a rare condition, and few cases in children have been reported. The sinus usually passes from the base of the penis near the abdominal wall toward the anterior bladder without communicating with the urinary tract.<sup>6</sup> The diagnosis is generally established by confirming the presence of the sinus tract using fistulography or the US. Magnetic resonance imaging may also be useful. The diagnosis in our case was

confirmed with US, and fistulography revealed the precise anatomy of the sinus.

Variants of CPS have been reported, and because the anatomic features often differ, a consensus concerning the embryology and classification has not yet been established. Rozanski et al<sup>7</sup> reported a case of CPS that represented a mild form of a midline closure defect. A second theory is that the defect does not allow for complete replacement of the ventral cloacal membrane by the lateral mesodermic folds, creating a fistulous tract.<sup>8</sup> van der Putte<sup>9</sup> considered the basis for this condition to be congenital malformations of cloaca-derived orifices, such as hypospadias, epispadias, vesical and cloacal exstrophy, double urethra, and prepubic sinuses. Most reported cases of CPS have been characterized by a tiny opening in the prepubic area that was primarily surrounded by squamous epithelium. Campbell et al<sup>10</sup> reported three cases of CPS in which the sinus tract was surrounded by stratified squamous epithelium or transitional epithelium with concentric bundles of collagen and smooth muscle fibers. The authors concluded that these sinuses may represent a variant of epispadiac duplication of the urethra. Huang et al<sup>1</sup> performed an immunohistochemical study, and found that the epithelium lining was transitional proximally and squamous distally in five patients. They reinforced the theory that CPS is a variant form of dorsal urethral duplication. In our case, the squamous epithelium was surrounded by muscle and glands within the tissue, and urothelial epithelium was found within the sinus tract. The difference between the locations of the squamous epithelium and transitional epithelium may have been because of the fact that CPS reacts with ductal and glandular epithelia, including the urothelium. This finding might support the theory that dorsal urethral duplication is the etiology of this condition. Stephens<sup>11</sup> stated that CPS might be caused by an inturned epithelial tube or remnant along the line of fusion of the infraumbilical abdominal walls, and classified this into three parts based on where the sinus begins. We believe that this case involved type 2 dorsal urethral duplication



**Fig. 2** Pathologic specimen of dorsal urethral duplication.

according to Stephens' classification, which describes a sinus that simulates an accessory urethra and tract from the dorsum of the penis behind the symphysis pubis to the bladder wall. However, we found no previous studies indicating that the sinus continues with the urinary tract tissue mimicking the corpus spongiosum. It is possible that the sinus tract was hypertrophic because of the accumulation of discharge from the CPS.

In conclusion, the embryological explanation of the development of CPS is unclear. We postulate that all the previously reported cases may be manifestations of a wide spectrum of the same condition. However, according to most investigators, CPS may represent a variant of dorsal duplication of the urethra. To the best of our knowledge, this is the first case in which the CPS continued outward from the skin within the urinary tract. Complete excision is the most definitive form of treatment.

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