



## Urethral duplication presenting as a prepubic sinus in a male: A case report

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### ABSTRACT

**Background:** Congenital prepubic sinus (CPS) is a rare congenital variant of urethral duplication, with less than 50 cases documented in the scientific literature since it was first reported by Campbell et al., in 1987.

**Case presentation:** An 8-year-old male patient complained of an inflammatory secretion from a sinus opening on the dorsal base of his penis. Physical examination revealed an accessory orifice in the midline of the dorsal proximal penis without any penile curvature. An ultrasound (US) examination found an 8-mm collection of cutaneous and subcutaneous tissue at the dorsal base of the penis. The renal US, however, showed no abnormalities. Additionally, micturating cystourethrogram (MCUG), was negative for vesicoureteral reflux (VUR). Retrograde sinusography (RS) and urethrogram showed no communication between the urinary tract, and the sinus was found to end blindly at the level of the pubic symphysis. A surgical excision was decided, to avoid potential future complications. A 3-cm-long intact sinus was dissected and excised entirely. Histological examination confirmed the diagnosis of a urethral duplication. The postoperative course was uneventful. To date, at six-month of follow-up, the patient remains asymptomatic.

**Conclusions:** The histological findings in this case support the theory that CPS is a variant of dorsal urethra. Urethral duplication should always be suspected in the case of accessory dorsal openings. Surgical excision is recommended at an early age, even in the absence of symptoms, to improve aesthetics, prevent recurrent infections and avoid the development of future malignant tumors.

### 1. Introduction

Urethral duplication is a rare congenital malformation with only around 300 reported cases in scientific literature [1]. It can manifest in various forms, and the rarest variant is the congenital prepubic sinus (type 3 urethral duplication according to Stephens' classification), with less than 50 cases reported in the literature since 1987 [2–4]. Congenital prepubic sinus (CPS) is a congenital anomaly characterised by a small tract originating in the skin overlying the symphysis pubic, superior to the base of the penis or clitoris, and extending toward the anterior bladder wall and the pelvic urethra, without direct communication with them [5–8]. Due to the rarity

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of this pathology, we aim to share our experience and review the literature to improve the scientific understanding of this condition (see Table 1).

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## 2. Case presentation

An 8-year-old male patient was admitted to our Pediatric Emergency Department with complaints of intermittent mucous and purulent leakage from an orifice located at the dorsal base of the penis.

The patient had no history of fever, urinary symptoms or familiar history of urinary anomalies. Hematologic exams, urine exams and urine culture test results were all negative.

During physical examination, the testicles were observed to be normal, the penis was well-developed with a urethral meatus at the tip of the glans and a sinus at the dorsal base of the penis surrounded by normochromic skin (Fig. 1). An ultrasound examination (US) revealed an 8 mm cutaneous and subcutaneous tissue collection at the dorsal base of the penis. Renal ultrasound was normal. Micrurating cystourethrogram (MCUG) was negative for vesicoureteral reflux (VUR). We conducted a retrograde sinogram (RS) and urethrogram by injecting a contrast agent: these exams showed no communication between the urinary tract and the sinus which ended blindly at the pubic symphysis (Fig. 2).

Following 10 days of antibiotic treatment (Cefixime) for the purulent discharge, surgery was arranged. Under general anaesthesia, an erection test was conducted yielding normal results. The sinus was marked and methylthioninium chloride was injected into the sinus; as a result, a 3 cm-long sinus tract extending superiorly towards the pubic symphysis was identified and completely excised. (Fig. 3 a, b, c). A vesical catheter was left in place for one day. There were no complications observed during or after the surgery. The post-operative course was uneventful. Histological examination confirmed the clinical diagnosis of congenital prepubic sinus (Stephens type 3 dorsal urethral duplication) (Fig. 4 a,b,c). After a six-month follow-up, the patient has not experienced any symptoms to date.

## 3. Discussion

Urethral duplication (UD) is a rare urological malformation in pediatric age [9]. Although the true incidence of the condition remains unknown, it has been observed that males are more frequently affected [10]. Urethral duplication can occur in either the coronal or sagittal planes, resulting in dorsal and ventral urethra [9–13]. It can be an isolated condition or it may present in association with other genitourinary anomalies such as hypospadias or, even more rarely, epispadias [13]. Stephens' classification identifies three types of dorsal urethral duplication: type 1 is a complete or incomplete tandem channel that follows the normal urethra from the bladder to the glans and which may join the urethra at some point or may end blindly; type 2 is an epispadiac type of channel from the dorsum of the penis to the bladder, which may or may not join the urethra at some point; type 3 (congenital dermoid sinus or congenital prepubic sinus) is a dermoid tubular structure with variable histological findings that begins in a tiny opening of the skin overlying the pubic symphysis and can course above, below or, in only a very few cases, directly through the pubis [2,5,14].

Our patient's case corresponds to type 3 of Stephens's Classification in terms of anatomical features and histological findings.

The exact embryological origin of the congenital prepubic sinus is still unclear and four distinct theories have been proposed [2,14].

Leakage from the sinus is the most common symptom, as reported in the literature, and our patient presented to our attention with the same complaint.

In line with the literature, he underwent an intraoperative erection test, which showed an absence of penile curvature; in fact, congenital penile curvature was found in only 3 out of 28 male patients (11 % prevalence) [15,16].

Penile curvature associated with some cases of CPS may be evaluated through an erection test to determine the presence/extent of the curvature. Surgical correction may be considered if needed [17].

Since the sinus simulating the accessory urethra in CPS opens on the dorsal side of the penis, CPS can be defined as a variant of epispadiac dorsal urethral duplication [5].

The features of penile epispadias include a dorsally grooved glans, minimal external rotation of the corpora cavernosa, slight shortening and dorsal curvature of the penis' shaft and divergence of corpora cavernosa. Penile curvature can be caused by abnormal development of the urethral plate and corporal disproportion or differential growth of normal corpora cavernosa [18].

Our patient's clinical onset age (8 years old) is older than the median age reported in the literature (2 years old) [19]. In our case, we performed US, MCUG and RS, which are widely used in medical literature. Although the use of computed tomography (CT) is also mentioned in the literature, we opted not to use CT scans: since RS reported the obliteration of the tract and the absence of communication with the urinary system, we determined that a CT scan would not have provided any useful pre-operative information. Therefore, we chose to prioritize the patient's safety and avoid any unnecessary radiation exposure by opting not to perform the scan and proceeding with the surgical procedure as planned [20]. US, MCUG and RS can give a more precise picture of the anomaly: US can be used to evaluate the presence of associated genitourinary malformations that usually occur with urethral duplication (ureteropelvic junction obstruction, renal ectopia or agenesis, bladder duplication, posterior urethral valves). MCUG is used to show potential communication between the bladder, the functional urethra and the sinus, reveal the functional urethra and show the presence of VUR, which is associated with UD in 37.5 % of cases [11]. RS is pivotal for determining the precise relation of the sinus to other pelvic structures. As demonstrated by our case, surgical intervention is a necessary and effective treatment to prevent recurrent infections, improve aesthetics and avoid the reported development of malignant tumors [21,22].

**Table 1**  
Literature review.

Author	N. patients	Age/median age at diagnosis	Sex	Signs and symptoms	Other congenital anomalies	Imaging	Treatment	Histopathology
Lei B.Z et al., 2020	1	18 years	M	Pubic tubular structure draining to the skin	\	Uro - MRI	Surgical excision	Urothelium, stratified columnar epithelium and squamous mucosa
Mostafa M. S et al., 2019	1	2 years	M	Suprapubic asymptomatic sinus	\	US MCUG	Surgical excision	Squamous epithelium lining the sinus with no evidence of transitional or urothelial epithelium.
K.M.S Al-Wattar; 2003	1	5 years	M	Pre-pubic tubular structure without secretions	\	US Uro-MRI	Complete surgical excision	Proximal transition like epithelium and a squamous epithelium at the external end. Presence of smooth muscle bundles and connective tissue.
Campbell J. et al., 1987	3	11 months	1 M 2F	Inflamed supra-pubis midline opening and bilateral inguinal lymphodanopathy	\	Retrograde sonography	Surgical excision	Transitional epithelium. The sinuses was surrounded by bundles of smooth muscle
Fukuhara M et al., 2021	1	13 years	M	Fistula opening near the dorsal penis	\	Renal US CT	Extraperitoneal excision	Stratified squamous and transitional epithelial lining.
Soares-Oliveira M.; 2002	2	6.5 months	2 M	Chronic discharge from a prepubic midline orifice	\	Renal US Retrograde Sonography MCUG	Surgical excision	Stratified squamous epithelial lining.
Montenero J.A et al., 2022	1	2 years	M	Prepubic fistulous orifice	\	Renal US Retrograde sonography Cystoscopy	Surgical complete excision	Low and high molecular weight keratins and a transitional pattern for keratin 7 and GATA3, with positivity at <i>cul de sac</i> level and negativity at proximal level.
Aeron R. et al., 2017	1	20 years	M	Intermittent mucous discharge from a prepubic midline opening	Penile curvature	Sinogram Retrograde urethrogram	Exploration and circumcoronal excision	Stratified squamous epithelium distally with urothelium lining and few smooth muscle bundles
Huang C.C et al., 2001	5	2 years [1 month-14 years]	3 M 2F	Purulent discharge form the sinus opening	\	Sinogram	Surgical excision	Transitional and squamous epithelium lining the sinus, smooth muscle bundles around the sinus tract
Sasaki Y et al., 2010	1	4 months	M	Discharge from the prepubic sinus opening.	\	Fistulography Uro-RMI	Complete surgical excision	Stratified squamous, transitional, and cylindrical epithelial cells
Wang C. et al., 2019	1	10 years	M	Pain in the dorsal base of the penis and occasional discharge from the sinus	Penile curvature	Retrograde sinogram Retrograde urethrogram CT	Complete surgical excision and correction of the penile curvature	Transitional epithelium inside the sinus and a few smooth muscle bundles identified around the sinus
Guler Y. et al., 2019	1	4 years	M	Purulent discharge from the distal part of the dorsum of his penis	\	Fistulography	Complete surgical excision	Multiple lamellar squamous epithelium
Nazir Z. et al., 2019	4	9 months 18 months 2 years 13 years	1 M 3 F	Purulent discharge from tiny opening at pubis	\	US MCUG Cystoscopy Sinogram	Complete surgical excision	Admixture of stratified squamos, columnar and transitional epithelium surrounded by smooth muscle fibers and inflamed fibrous tissue.
Duman L. et al., 2017	1	20 months	M	Recurrent purulent discharge from opening located over the prepubic area	\	US Uro-MRI Retrograde urethrogram	Complete surgical excision	Stratified squamous epithelium and surrounded by bundles of smooth muscles

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Table 1 (continued)

Author	N. patients	Age/median age at diagnosis	Sex	Signs and symptoms	Other congenital anomalies	Imaging	Treatment	Histopathology
Eroglu E. et al., 2016	1	7 years	M	An erythematous skin lesion was noticed at the dorsal aspect of penis	\	Postoperative urinary system US	Complete surgical excision	Uroepithelium
Celebi S. et al., 2014	1	3 years	M	Yellowish discharge from a tiny opening on the dorsal base of the penis	\	US Fistulography	Complete surgical excision	Squamous epithelium and continued along the sinus tract, which was lined with urothelial epithelium
Kobayashi H. et al., 2015	1	22 years	F	High grade fever and lower abdominal pain;	\	US CT Cystoscopy Sinogram	Complete surgical excision	Urothelial cell layer on the bladder side, while revealing a squamous cell layer on the open side.
Shaw J.S. et al., 2015	1	16 years	F	Clitoral and left labial swelling and pain.	\	US MRI Cystoscopy Fistulography	Surgical excision combining vaginal and laparoscopic approaches.	Squamous epithelium and transitional epithelium with underlying smooth muscle
Yamada K. et al., 2012	1	10 months	F	Purulent discharge from a small pinhole in her lower abdomen	Urachal remnant	MRI Sinogram	Complete surgical excision	Stratified squamous epithelium near the skin side and cystic part, but transitional epithelium was present at the end,
Nasir A. et al., 2012	1	9 months	M	Recurrent muco-purulent discharge from a tiny opening on the base of the penis.	Ventrally hooded prepuce, dorsal chordee, penile torsion to the left	MCUG	Complete surgical excision and correction of penile curvature	Transitional epithelium
Ozdemir E. et al., 2011	1	4 years	M	Mucopurulent discharge at the proximal dorsum of the penis.	\	Sinogram	Complete surgical excision	Squamous epithelium.
Hayase M. et al., 2006	1	12 years	F	Clitoromegaly and a sinus on the midline prepubic area	Clitorio-megaly	MRI Retrograde urethrography Cystoscopy Sinogram	Complete surgical excision	Squamous epithelium.
Usami M. et al., 2005	1	3 years	M	Yellowish discharge from a tiny opening in the midline prepubic area on the dorsal base of the penis.	\	\	Complete surgical excision	Stratified squamous epithelium and surrounded by bundles of smooth muscle and collagen
Samujh R. et al., 2004	1	8 years	M	Discharge from an opening overlying the pubic symphysis on the dorsal base of the penis.	\	MCUG Sinogram	Complete surgical excision	Stratified squamous epithelium and bundles of smooth muscle
Tsukamoto K. et al., 2004	2	3 months 4 years	1 M 1F	Opening in the midline prepubic area	\	Sinogram	Complete surgical excision	Squamous and transitional epithelium; transitional epithelium and surrounded by bundles of smooth muscle fibers.
Balster S. et al., 2003	1	2 years	M	Discharge from an opening overlying the pubic symphysis on the dorsal base of the penis.	\	Sinogram	Complete surgical excision	Transitional and squamous epithelium
Jouini R. et al., 2003	1	3 months	M	Muco-purulent discharge from a tiny opening on the base of the penis.	\	Urethrography Fistulography.	Complete surgical excision	Transitional epithelium and surrounded by bundles of smooth muscle fibers.
Ergun O. et al., 1998	3	10 months 4 years 5 years	M	Muco-purulent discharge from an opening on the base of the penis.	\	US MCUG Sinogram	Complete surgical excision	Stratified squamous epithelium
Green J.S.A. et al., 1997	1	10 months	F	Pustule in labial folds anterior to the clitoris	\	Cystoscopy Sinogram	Complete surgical excision	Stratified squamous epithelium
Daher P. et al., 1994	1	2 months	F	Yellowish discharge from a tiny opening 2cm above the clitoris facing the pubic symphysis	\	MCUG Urethrogram Fistulography	Complete surgical excision	Stratified squamous epithelium

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Table 1 (continued)

Author	N. patients	Age/median age at diagnosis	Sex	Signs and symptoms	Other congenital anomalies	Imaging	Treatment	Histopathology
Park W.H. et al., 1993	1	4 months	F	Yellowish discharge from a tiny opening in the midline prepubic area immediately superior to the base of the clitoris	\	US Sinogram	Complete surgical excision	Stratified squamous epithelium and surrounded by concentric bundles of collagen and smooth muscle with mild inflammatory infiltrates
Gonzalez J. et al., 1993	1	1 year	M	Accessory orifice on the base of the penis.	\	US MCUG Retrograde urethrography	Complete surgical excision	Stratified squamous epithelium
Rozanski T.A. et al., 1990	1	10 months	F	Discharge from an opening overlying the pubic symphysis	\	US Retrograde cystogram	Complete surgical excision	Squamous epithelium-lined sinus tract.

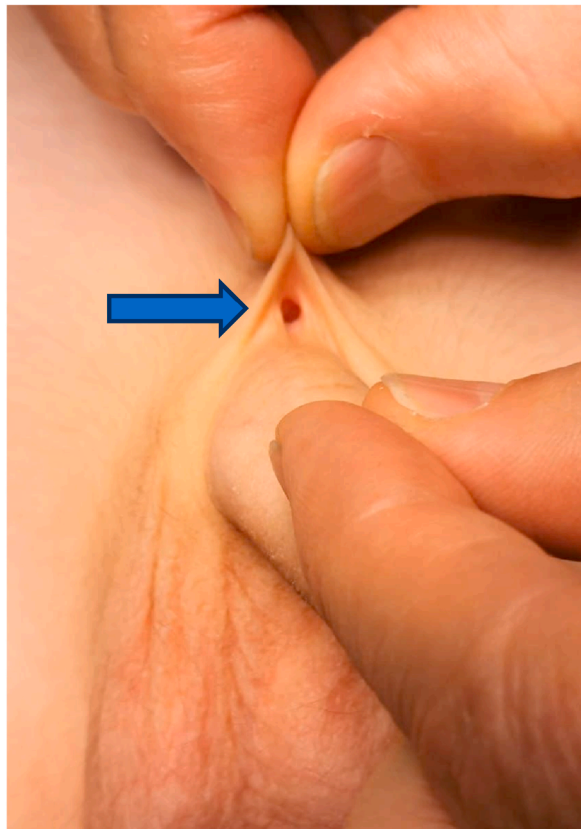


Fig. 1. Sinus at the dorsal base of the penis with surrounding normochromic skin.

#### 4. Conclusion

Congenital prepubic sinus is a rare subtype of dorsal urethral duplication that lacks clear diagnostic and treatment protocols in the current medical literature. A cutaneous sinus at the base of the penis with secretions, even alongside a normal penis, should be thoroughly evaluated to confirm or rule out a CPS. Surgical excision of CPS is recommended at an early age, even in the absence of symptoms, to improve aesthetics, prevent recurrent infections and avoid the development of malignant tumors.

#### Consent for publication

Informed written consent was obtained from the parents of the child described in this study to publish the clinical data and photographs.

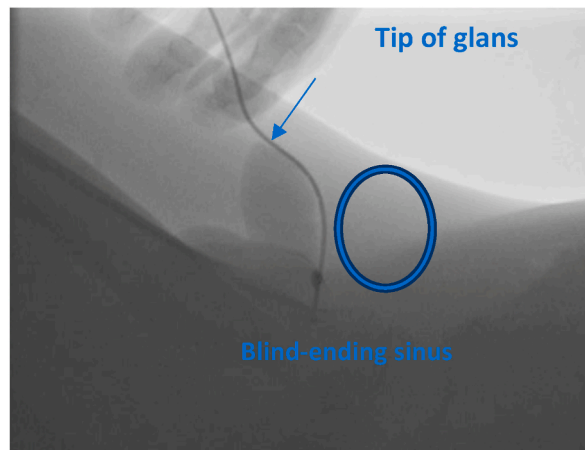


Fig. 2. Retrograde sinogram after injection of contrast agent does not show any communication between the urinary tract and the sinus which ends blindly at the level of the pubic symphysis.

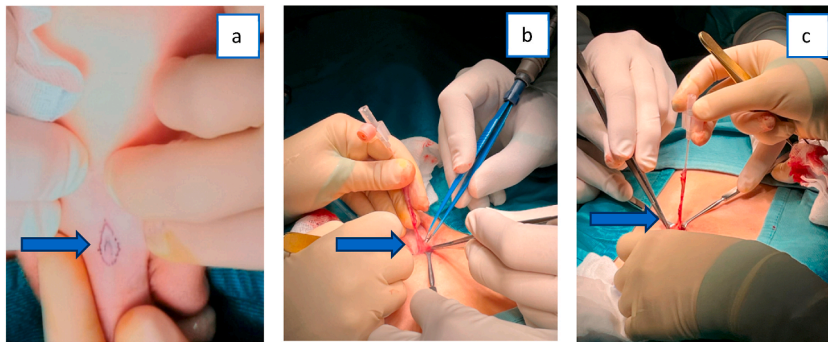


Fig. 3. a Marked orifice at the dorsal base of the penis with surrounding normochromic skin. 3b,c Surgical correction by excision of a 3 cm-long-sinus tract.

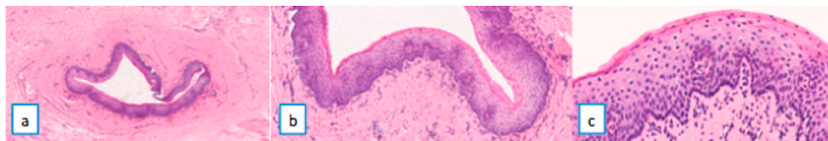


Fig. 4. a,b,c Sinus was lined by stratified squamous epithelium and surrounded by smooth muscle and collagen bundles at different power of magnification. a) 3 × OM, Hematoxylin and Eosin. b) 9 ×, OM, Hematoxylin and Eosin. c) 22 ×, OM, Hematoxylin and Eosin.

**Authors statement**

During the preparation of this work the authors did not use any specific tools or service, they reviewed and edited the content as needed and take full responsibility for the content of the publication.

**Informed consent**

Written informed consent was obtained from the parents of the minor patient, who are the legal guardian of the baby, for the publication of any potentially identifiable images or data included in this article.

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**Authorship**

All authors attest that they meet the current ICMJE criteria for authorship.

## Author contributions

FN and MM drafted the initial manuscript, reviewed, and revised the manuscript and conducted the review of the literature. RA and MM conceived the idea for this case report, coordinated and supervised data collection and the research group, and critically reviewed the manuscript. FM and RA contributed to the diagnostic and therapeutic management of the baby before and after birth, as well as contributing to the acquisition and interpretation of the data. All authors contributed to the article and approved the submitted version.

## Declaration of competing interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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## List of abbreviations

CPS	congenital prepubic sinus
UD	urinary duplication
US	Ultrasound
MCUG	Micturing cysto-urethrogram
VUR	vesicoureteral reflux
RS	retrograde sinusography
Uro-MRI	Uro-magnetic resonance imaging
CDS	Congenital dermoid sinus

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