



A Rare Case of Female Congenital Prepubic Sinus

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Abstract

Congenital Prepubic Sinus (CPS) is an uncommon urogenital anomaly. Our case is a 2 years old girl was diagnosed suspected prepubic sinus when she was 8 months old. With the methylene blue injection via the external opening, it was confirmed that the tract communicated with urinary bladder dome under cystoscopy. After reviewing the recent English literature, our case may be the first one which was proved fistula with direct vision. Our case may be a new disease or a new type of prepubic sinus.

Introduction

Congenital Prepubic Sinus (CPS) is a rare urogenital anomaly. It is characterized by an epithelial lined tract, which occasionally extends from skin over the pubis close to the penile root or clitoris to urinary bladder, urethra or umbilicus. The common complaint is the tiny cutaneous opening over suprapubic and prepubic skin with watery or purulent discharge with cutaneous abnormality because of local skin insult by secretions after the first report by Campbell et al. about 50 cases are reported in literature [1-8].

This is the first case, undergoing surgical management in our hospital. We clearly outlined the entire fistula tract and discovered an inner opening in urinary bladder dome, which is never found in previous reports.

Case Presentation

A 3-month-old girl presented to our office with a cutaneous opening superior to the urethra and the clitoris (Figure 1). Initially, epispadias or urethral duplication was suspected, and the Voiding Cystourethrography (VCUG) was performed, revealing that there was no connection between the cutaneous opening and her urethra. The sonography was performed and there was negative finding of bilateral kidneys and urinary bladder. Fistulogram was arranged when the patient was 8-month-old; the result was a deep and curvy tract (Figure 2) because there was no evidence of vesicocutaneous fistula, urethral duplication was suspected.

The patient was under follow-up in the office. The patient was taken back to our office because patient was found urine leakage from the opening when she was 2-year-old. The cystoscopy was arranged. During the pathway from the urethra into the bladder, there was no any fistula from urethra to bladder neck. A 24 Fr. Catheter was cannulated through the cutaneous opening and methylene blue was injected. Methylene blue leaked *via* a pinhole over the bladder dome (Figure 3) for better evaluation of the fistula, pelvic Computed Tomography (CT) was scheduled. The findings of CT were separated symphysis pubis and the fistula located above the urinary bladder (Figure 4). Therefore, the surgical intervention was arranged for the patient. A transverse incision was made over the lower abdomen to approach the urinary bladder. The urinary bladder was identified, and a tubular structure was noted at mid-line and connected to the urinary bladder dome (Figure 5) the methylene blue was injected from the external opening, and the bladder was opened to confirm and localize the internal opening of the fistula. The distal end of fistula was found passing a central bony defect of symphysis pubis. The distal end was incised at the level of pubic symphysis. The proximal end of the fistula was incised at the level of bladder dome. The fistula was resected, and the urinary bladder was closed.

The pathological report of the specimen was transitional epithelia. The girl is doing well later to the operation.

OPEN ACCESS

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Received Date: 05 Apr 2021

Accepted Date: 10 May 2021

Published Date: 14 May 2021

Citation:

Liao FT, Lin GB, Chen MF, Chia ST.
A Rare Case of Female Congenital
Prepubic Sinus. *Clin Surg.* 2021; 6:
3164.

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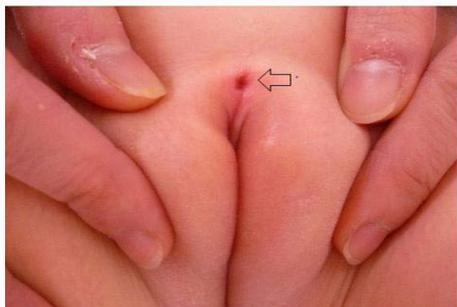


Figure 1: Clinical picture shows a cutaneous opening superior to the urethra and the clitoris.



Figure 2: Fistulogram shows a deep and curvy fistula without connecting to urinary bladder and bowel loops.

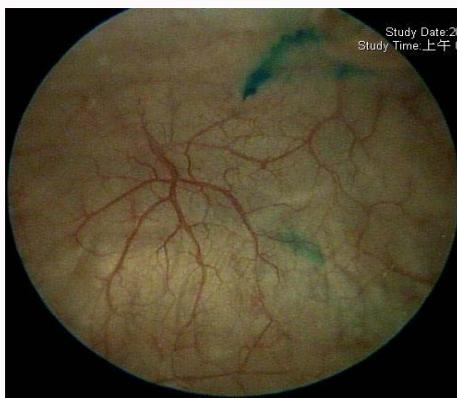


Figure 3: Cystoscopy shows methylene blue is found at bladder dome when dye was injected from cutaneous opening.

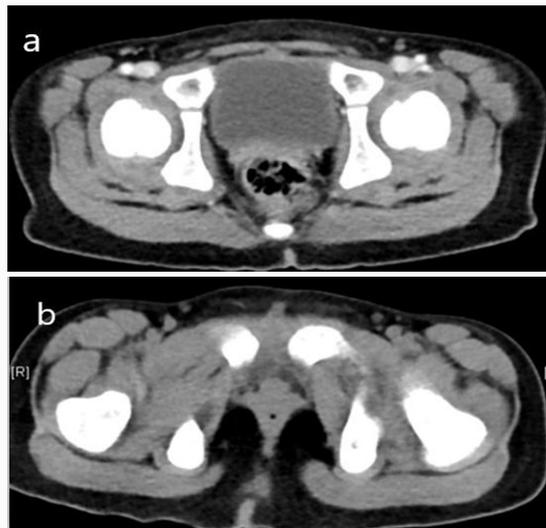


Figure 4: Pelvic CT shows a) a tubular structure near urinary bladder b) widening of pubic symphysis.



Figure 5: View of operation. a) A fistula connected to urinary bladder dome with external opening cut, b) the fistula.

Discussion

CPS occasionally presents in infancy and young children (preponderance in females) with discharging sinus in pubic area, which is close to the penile or clitoris [1]. CPS is also reported in literature as suprapubic fistula, suprapubic dermoid sinus etc. [3,4]. There are usually no obvious abnormalities at ultrasonography of urinary tract, voiding cystourethrogram and cystoscopy. Conventional fistulography and fine lacrimal probe or plastic sheath of intravenous cannula may play a role to delineate the anatomy [3].

Since Campbell et al. published the 1st case in 1987, there are around 36 reports about CPS in the English literature (51 cases; Table 1, 2), which include 27 males and 24 females, aging from 1 month to 22 years old [1-4]. The most common clinical manifestation of CPS

is the discharge from a midline opening between the dorsal penile root or clitoris and the prepubic region in infancy from the table. The prepubic sinus tracts were above the pubis in 16 cases, through the pubic symphysis in 7 cases, and below the pubis in 9 cases. The proximal end of the sinus tract attached to the urinary bladder in 29 cases, the umbilicus in 4 cases, the urethra in 2 cases, the retropubic space in 6 cases, the pubic symphysis in 5 cases, and the prepubic space in 3 cases. The histological results of the tracts were pure squamous or stratified squamous epithelium was reported in 16 cases, pure transitional epithelium in 7 cases, mixed squamous/stratified squamous and transitional epithelium in 18 cases, and cylindrical or columnar epithelium in 4 cases (Table 3).

For our case, it should be focused on the internal opening of this

Table 1: Case report of CPS in the English literatures.

Case report of CPS in the English literatures							
Year	Age	Gender	Symptom	Cutaneous opening	Tract	Distal opening	Histology(epithelium)
1987	4M	F	Discharge	Prepubis	Above pubis	Urinary bladder	Proximal transitional, distal squamous
	6M	M	Opening	Prepubis	Above pubis	Urinary bladder	Stratified squamous
	2Y	F	Groin swelling	Above clitoris	Above pubis	Urinary bladder	Proximal transitional, distal squamous
1989	18M	F	Discharge	Suprapubis	Below pubis	Urinary bladder	Transitional
1990	10M	F	Discharge	Prepubis	Above pubis	Urinary bladder	Squamous
1992	2M	M	Polypoid opening	Prepubis	Through pubic symphysis	Urinary bladder	Transitional
	2Y	F	Discharge	Prepubis	Above pubis	Urinary bladder	Pseudostratified
	2Y	F	Opening	Prepubis	Above pubis	Retropubic space	Transitional
1993	4M	F	Discharge	Above clitoris	Above pubis	Umbilicus	Proximal transitional, distal squamous
1994	11M	M	Discharge	Suprapubis	Above pubis	Urinary bladder	Proximal transitional, distal squamous
	3Y	M	Inflammation red& swelling	Prepubis	Above pubis	Urinary bladder	Proximal transitional, distal squamous
	2M	F	Discharge	Above clitoris	Through pubic symphysis	Urinary bladder	Stratified squamous
1995	1Y	F	Discharge	Above clitoris	No detail	Retropubic space	Squamous, transitional, columnar
1996	8Y	F	Opening	Above clitoris	Below pubis	Retropubic space	Squamous and transitional
1997	10M	F	Pus	Above clitoris	Below pubis	Urethra	Stratified squamous
	10M	M	Discharge	Penile dorsal radix	Above pubis	Urinary bladder	Stratified squamous, cylindrical
	5Y	M	Discharge	Prepubis	Above pubis	Urinary bladder	Squamous
	4Y	M	Discharge	Dorsal penile root	Above pubis	Urinary bladder	Stratified squamous
1998	5Y	M	Discharge	Suprapubis	No detail	Abdominal wall	Squamous, columnar
2001	2M	F	Discharge	Above clitoris	No detail	Urinary bladder	Proximal transitional, distal squamous
	3M	M	Discharge	Prepubis	No detail	Urinary bladder	Proximal transitional, distal squamous
	1M	M	Discharge	Prepubis	No detail	Urinary bladder	Proximal transitional, distal squamous
	14Y	M	Discharge	Dorsal penile root	No detail	Prepubic space	Proximal transitional, distal squamous
	2M	F	Discharge	Prepubis	No detail	Dorsal tunica albuginea clitoris	Squamous
2002	8M	M	Discharge	Prepubis	Below pubis	Prostatic urethra	Stratified squamous
	5M	M	Discharge	Prepubis	No detail	Pubic symphysis	Stratified squamous
	5Y	M	Discharge	Prepubis	No detail	Retropubic space	Proximal ciliated columnar, middle transitional, distal stratified squamous
2003	4Y	F	Discharge	Suprapubis	No detail	Umbilicus	Transitional
	2Y	M	Discharge	Penile dorsal radix	No detail	Urinary bladder	Proximal transitional, distal squamous
2004	3M	M	Opening	Prepubis	Through pubic symphysis	Umbilicus	Proximal transitional, distal squamous
	4Y	F	No detail	No detail	No detail	Urinary bladder	Transitional
	8Y	M	Discharge	Penile dorsal radix	Above pubis	Urinary bladder	Stratified squamous
2005	3Y	M	Discharge	Penile dorsal radix	No detail	Pubic symphysis	Stratified squamous
2006	12Y	F	Clitromegaly	Above clitoris	No detail	Retropubic space	Squamous
2007	3Y	M	Discharge	Penile dorsal radix	Above pubis	Urinary bladder	Proximal urethral, distal squamous
2010	4M	M	Discharge	Prepubis	Above pubis	Urinary bladder	Stratified squamous, transitional, cylindrical
2011	4Y	M	Discharge	Penile dorsal radix	No detail	Prepubic space	Squamous
2013	9M	M	Discharge	Penile dorsal radix	Below pubis	Urinary bladder	Transitional
	10M	F	Discharge	Prepubis	Through pubic symphysis	Urinary bladder	Proximal transitional, distal stratified squamous
2015	16Y	F	Clitoral and left labial swelling	No detail	Through pubic symphysis	Urinary bladder	Proximal transitional, distal squamous
	22Y	F	Fever, abdominal pain	Prepubis	Below pubis	Urinary bladder	Proximal urethral, distal squamous
	3Y	M	Discharge	Prepubis	Below pubis	Prepubic space	Proximal urethral, distal squamous

2016	2Y	F	Discharge	Prepubis	Above pubis	Umbilicus	Proximal urethral, distal squamous
	8M	M	Discharge	Penile dorsal radix	No detail	Pubic symphysis	Squamous
2017	20Y	M	Discharge	Penile dorsal radix	Through pubic symphysis	Pubic symphysis	Stratified squamous
2019	9M	F	Discharge	Above clitoris	Below pubis	Urinary bladder	Proximal transitional, distal stratified squamous
	2Y	M	Pus	Penile dorsal radix	Below pubis	Urinary bladder	Stratified squamous, columnar, transitional
	13Y	F	Labial abscess	Above clitoris	No detail	Retropubic space	Stratified squamous, columnar, transitional
	1M	F	Discharge	Prepubis	No detail	Urinary bladder	Stratified squamous, columnar, transitional
	4Y	M	Pus	Penile dorsal radix	No detail	Pubic symphysis	Squamous
This case	2y	F	Discharge	Prepubis	Through pubic symphysis	Urinary bladder	Transitional

Table 2: Patient demographic of CPS in the English literatures.

All patients	51
Sinus	49
Fistula	2
Age (median+SD)	2-264 m/o(24 ± 62)
Gender	
Male	27
Female	24

: one case is our case, and another is revealed in MRI image [8]

tract. There are no similar cases in recent literatures that the real opening is located in urinary bladder. Indeed, in our case, it is at the urinary bladder dome.

A few embryological hypotheses are developed to explain the pathogenesis of CPS. The 1st hypothesis is a localized failure of midline fusion in the lower abdominal wall [1]. It is reported that some cases of CPS are accompanied with diastasis of the pubic symphysis [1]. The hypothesis may be supported when there is stratified squamous epithelium in the entire tract.

The 2nd hypothesis considers the prepubic sinus as a variant of dorsal urethral duplication [1]. Shaw JS et al. have a figure for urethral duplication (Figure 2A). However, in our case, the tract communicated with urinary bladder dome, which may be incompatible to the definition of urethral duplication. Therefore, prepubic fistula or a new classification of prepubic sinus may be used for our case.

The 3rd hypothesis thinks CPS as a congenital fistula of the primitive urogenital sinus. CPS is classified into 3 anatomical types by Soares-Oliveira et al. high, toward the urachal remnant; middle, to the bladder; and low, to the prostatic urethra [1].

The 4th hypothesis is that a residual cloacal membrane and umbilicophallic groove cause CPS. It's based on the finding of the proximal blind end of all prepubic sinuses [7]. Because there is a true communicating with bladder cavity in our case, the concept of residual cloaca membrane is not favored.

Complete and meticulous excision of prepubic sinus is essential to prevent recurrent symptoms, infections and risk of malignant transformation [5]. A fine lacrimal probe or an intravenous cannula can be helpful for complete excision [3].

In our case, the prepubic sinus was found in early infancy stage. The surgical intervention was performed in toddler stage before the infection in the tract, which may destroy the structure of the internal opening. The methylene blue was used to confirm the internal

Table 3: Data of prepubic sinus.

Clinical manifestation	
Discharge	36
Infection	6
Swelling lesion	3
Tract	
Above pubis	16
Below pubis	9
Through pubic symphysis	7
Cutaneous opening	
Prepubis	22
Above clitoris	10
Penile dorsal radix	13
Suprapubis	4
Distal end	
Urinary bladder	29
Retropubic space	6
Prepubic space	3
Pubic symphysis	5
Umbilicus	4
Urethra	2
Abdominal wall	1
Dorsal tunia albuginea clitoris	1
Histology of specimen	
Squamous epithelium	16
Transitional epithelium	7
Proximal transitional epithelium+ distal squamous epithelium	19

opening of the tract was in urinary bladder under cystoscopy. The internal opening is so tiny; without methylene blue, the internal opening is hardly detected.

There was no proven communication between prepubic sinus and urinary bladder or other structure in the previous reports [7]. It's not sure whether this is another type of prepubic sinus or a new disease in our case. Prepubic fistula may be a suitable nomenclature if more and more similar cases are reported.

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