Congenital prepubic sinus accompanied by prevesical abscess

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Abstract

Congenital prepubic sinus is an extremely rare anomaly. The etiology is uncertain and the anatomical features often differ from each other. We report a 22-year-old woman with a congenital prepubic sinus accompanied by a prevesical abscess. She was admitted to our hospital with high-grade fever and low abdominal pain. Computed tomography revealed a prevesical abscess. After treatment of the prevesical abscess, we completely excised the congenital prepubic sinus. To our knowledge, this is the first reported case that accompanied by prevesical abscess on a congenital prepubic sinus. Moreover, this case represents the oldest reported age of a patient with a congenital prepubic sinus.

Introduction

Congenital prepubic sinus is an extremely rare anomaly. The etiology is uncertain and the anatomical features often differ from each other. In most cases, the chief complaints of this anomaly were discharge from the opening sinus and received operation in childhood. Here, we present our experience of an adult patient with congenital prepubic sinus accompanied by prevesical abscess.

Case Report

A 22-year-old woman visited our hospital with high-grade fever and low abdominal pain while urinating. Physical examination revealed low abdominal tenderness and pus discharging from skin near the pubic region. Urinary findings and urinalysis were normal. Routine hematological and chemical evaluation revealed a high level of white blood-cells and C-reactive protein. Abdominal ultrasound revealed no residual urine, but detected an isoechoic lesion in her prevesical space. Computed tomography (CT) with contrast medium also clearly revealed the prevesical abscess (Figure 1). Although cystoscopical findings were relatively normal, the mucosa of the anterior wall was slightly reddish. During the first surgical procedure, yellowish tissue surrounded by a capsule in front of the bladder was found. We excised a portion of the inflammatory tissue and inserted a tube in this space for drainage. Pathological findings revealed fat necrosis with granulation surrounded by a cystic wall. Urachal remnants were not detected in the tissue. She was discharged from the hospital one week later after her general condition had improved. She was re-admitted three months later to undergo a second surgical procedure on the sinus. Before this radical operation, we examined the relationship between the sinus and urinary tract by using contrast medium from the catheter inserted to opening sinus. Fluoroscopy revealed that there is no connection between this congenital prepubic sinus (CPS) and lower urinary tract. Plain CT that performed immediately after this examination endorsed this fluoroscopic view (Figure 2). Thus the sinus was diagnosed as an extremely rare anomaly that called CPS. In the operating room, we first injected indigo carmine into the opening of the CPS, inserted a catheter (0.7 mm in diameter) into the same opening, and then reached into the anterior area of the bladder using fluoroscopic monitoring in order to confirm the diagnosis. Excision of the CPS was performed using this catheter while occasionally conducting intraoperative fluoroscopic monitoring during the operation. In this case, the CPS went toward the pubic symphysis about 2 cm horizontally, descended to the lower level of the pubic symphysis, ascended steeply toward the top of the symphysis, and then crossed over the Y-shaped gorge of the symphysis. The periosteum was preserved carefully and the sinus descended steeply again, toward the deep area, along the back of the pubic symphysis, and went toward the anterior area of the bladder. The CPS ended in this area. We excised the end of the sinus, along with surrounding tissue, and injected a solution of sodium chloride into the bladder to confirm the absence of leakage. The length of this excised sinus was about 7 cm. Pathological findings of the sinus revealed a urothelial cell layer on the bladder side, while revealing a squamous cell layer on the open side. The patient had an uneventful postoperative course.

Discussion

CPS is an extremely rare congenital anomaly of the external genitalia. To our knowledge, only 39 cases have been reported in the English literature since it was first described by Campbell *et al*.[1] in 1987 (Table 1).[1-25] While urethral duplication is a more often reported anomaly, CPS is extremely rare. The most important difference in these anomalies is that the latter is not communicating with the urinary tract. The etiology is uncertain and the anatomical features often differ from each other. We encountered this rare anomaly, and the anatomical findings in this case suggest new etiological findings of CPS. Although, in most reported cases, the route of the CPS is vague, especially near the pubic symphysis, the sinus in this case was extremely meandering as if to avoid the pubic symphysis. This means that the CPS existed in the early stage of fetal development, probably followed by the formation of the pubic symphysis. Pathological findings did not reveal urachal remnants after both the first and second surgeries. There is a possibility that formation of the pubic symphysis was incomplete because of this congenital anomaly, since the X-ray (KUB) of this patient revealed a loose symphysis pubis.

According to most reports in the English literature, CPS is considered a urethral developmental anomaly, a variant of dorsal urethral duplication. Three anatomical variants have been suggested by Stephens.[26] Type 1 is a complete or incomplete channel that runs close or

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parallel to the normal urethra from the glans to the bladder. Type 2 is an epispadiac channel that joins either the bladder or normal urethra. Type 3 is a dermoid sinus that simulates a urethra, but runs from the base of the penis or clitoris anterior to the pelvis, urethra, and bladder, behind the pubic symphysis to or towards the umbilicus.

There are five proposed theories for the etiology of CPS including the above theory. Rozanski *et al*.[3] proposed that CPS is a mild forme fruste of a midline abdominal wall closure defect. Chou *et al*.[9] proposed that CPS is a remnant of the cloaca. Soares-Oliveira *et al*.[15] proposed that CPS is a congenital fistula of the primitive urogenital sinus, with three anatom ic subtypes depending on the direction of the sinus tract: high, toward the urachal remnant; middle, toward the bladder; and low, toward the prostatic urethra. Tsukamoto *et al*.[19] proposed that CPS may be caused by a residual cloaca membrane and umbilicophallic groove, and that the depth may determine the position of the end of the sinus tract. Accordingly, with several different theories, the etiology of CPS still remains unclear.

Interestingly, in all reported CPS cases, there was a single surgical procedure, including exploration and excision. Only in this case was it necessary to perform two surgical procedures, owing to the treatment of the critical prevesical abscess. If she had not had a prevesical abscess, she probably would not have come to a hospital. Moreover, this case represents the oldest reported age of a patient with CPS. Although she had noticed pus discharging from her skin near the pubic region since childhood, she had no other signs or symptoms related to the genitourinary tract. This possibly

Table 1. Cases of congenital prepubic sinus reported in the English literatures.

Reference	Case No.	Sex	Age	Chief complaint
Campbell *et al*.[1]	1	Female	4 months	Discharge
	2	Male	6 months	Opening of prepubic sinus
	3	Female	2 years	Bilateral groin swelling
Crawford *et al*.[2]	4	Female	2 years	Opening of suprapubic sinus
Rozanski *et al*.[3]	5	Female	10 months	Irritated, inflamed, drained fluid
Lawson *et al*.[4]	6	Male	0 month	Polypoid opening of prepubic sinus
	7	Female	2 years	Discharge
	8	Female	0 month	Opening of prepubic sinus
Groff[5]	9	Female	0 month	Existence of a small bump
	10	Female	0 month	Opening of prepubic sinus
Park *et al*.[6]	11	Female	4 months	Discharge
Komura *et al*.[7]	12	Male	11 months	Inframed, discharge
	13	Male	3 years	Redness, swollen
Daher *et al*.[8]	14	Female	2 months	Discharge
Chou *et al*.[9]	15	Female	2 years	Discharge
Walther *et al*.[10]	16	Female	8 years	Enuresis
Green *et al*.[11]	17	Female	10 months	Pustule in her labial folds
Ergun *et al*.[12]	18	Male	10 months	Discharge
	19	Male	5 years	Discharge
	20	Male	4 years	Discharge
Nirasawa *et al*.[13]	21	Male	5 years	Opening of prepubic sinus
Huang *et al*.[14]	22	Female	2 months	Discharge
	23	Male	3 months	Discharge
	24	Male	1 month	Discharge
	25	Male	14 years	Discharge
	26	Female	2 months	Discharge
Soares-Oliveira *et al*.[15]	27	Male	8 months	Discharge
	28	Male	5 months	Discharge
Chao *et al*.[16]	29	Male	5 years	Discharge
Park *et al*.[17]	30	Female	4 years	Tiny grayish skin lesion and discharge
Al-Wattar[18]	31	Male	2 years	Discharge
Tsukamoto *et al*.[19]	32	Male	3 months	Opening of prepubic sinus
	33	Female	4 years	Opening of prepubic sinus
Usami *et al*.[20]	34	Male	3 years	Discharge
Hayase *et al*.[21]	35	Female	12 years	Clitoromegaly
Kim *et al*.[22]	36	Male	3 years	Discharge
Ozdemir *et al*.[23]	37	Male	4 years	Discharge
Nasir *et al*.[24]	38	Male	9 months	Discharge
Yamada *et al*.[25]	39	Female	10 months	Discharge
Present case	40	Female	22 years	High grade fever and abdominal pain

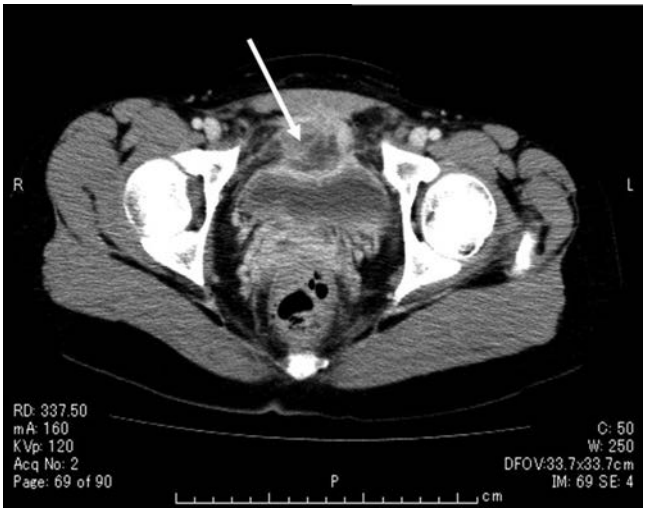

Figure 1. Computed tomography with contrast medium. White arrow indicates the abscess of perivesical space.

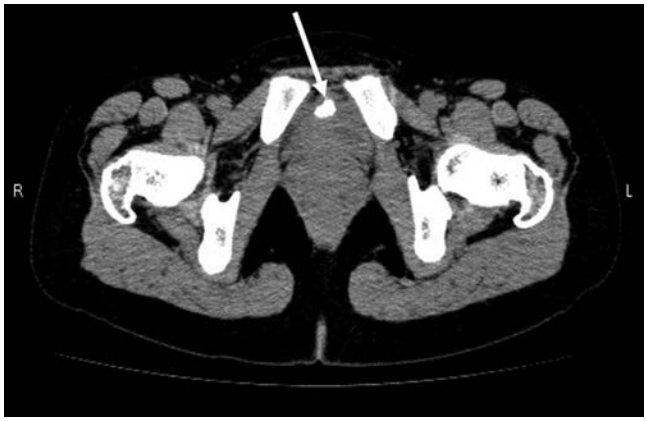

Figure 2. Computed tomography after injection of contrast medium from the prepubic sinus. White arrow indicates the sinus continuing near the anterior area of the bladder.

means that treatment of CPS is not necessarily required in all cases.

Conclusions

This case suggests the possibility that CPS is a latent congenital anomaly that is more prevalent throughout the world than expected. Otherwise, I propose another theory that neglected CPS for long time will cause the infections, like a prevesical abscess, sooner or later.

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