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 examination 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 gorse of the symphysis. The peristomum 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 (RUB) 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
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.5 proposed that CPS is a mild forme fruste of a midline abdominal wall closure defect. Chou et al.12 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.15 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

| 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.5 | 5 | Female | 10 months | Irritated, inflamed, drained fluid |
| Lawson et al.14 | 6 | Male   | 0 month  | Polypoid opening of prepubic sinus |
|           | 7 | Female | 2 years  | Discharge                       |
|           | 8 | Female | 0 month  | Opening of prepubic sinus       |
| Groff5    | 9 | Female | 0 month  | Existence of a small bump       |
|           | 10| Female | 0 month  | Opening of prepubic sinus       |
| Park et al.5 | 11| Female | 4 months | Discharge                       |
| Komura et al.1 | 12| Male   | 11 months | Inframed, discharge             |
|           | 13| Male   | 3 years  | Redness, swollen                |
| Daher et al.9 | 14| Female | 2 months | Discharge                       |
| Chou et al.12 | 15| Female | 2 years  | Discharge                       |
| Walther et al.15 | 16| Female | 8 years  | Enuresis                        |
| Green et al.16 | 17| Female | 10 months | Pustule in her labial folds     |
| Ergun et al.17 | 18| Male   | 10 months | Discharge                       |
|           | 19| Male   | 5 years  | Discharge                       |
|           | 20| Male   | 4 years  | Discharge                       |
| Nirasawa et al.11 | 21| Male   | 5 years  | Opening of prepubic sinus       |
| Huang et al.13 | 22| Female | 2 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.18 | 29| Male   | 5 years  | Discharge                       |
| Park et al.15 | 30| Female | 4 years  | Tiny grayish skin lesion and discharge |
| Al-Wattar19 | 31| Male   | 2 years  | Discharge                       |
| Tsukamoto et al.15 | 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 | Clitororomegaly                 |
| 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 |
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.

References

1. Campbell J, Beasley S, McMullin N, et al. Congenital prepubic sinus: possible variant of dorsal urethral duplication (stephens type 2). J Urol 1987;137:505-6.
2. Crawford RA, Sethia KK, Fawcett DP. An unusual presentation of a urachal remnant. Br J Urol 1989;64:315-6.
3. Rozanski TA, Kiesling VJ Jr, Tank ES. Congenital prepubic sinus. J Pediatr Surg 1990;25:1301.
4. Lawson A, Corkery JJ. Prepubic sinus: an unusual urachal remnant. Br J Surg 1992;79:573.
5. Groff DB. Suprapubic dermoid sinus. J Pediatr Surg 1993;28:242-3.
6. Park WH, Choi SO, Park KK, et al. Prepubic dermoid sinus: possible variant of dorsal urethral duplication (stephens type 3). J Pediatr Surg 1993;28:1610-1.
7. Komura J, Yano H, Kanazawa M, et al. Congenital prepubic sinus. Pediatr Surg Int 1994;9:287-9.
8. Daher P, Diab N, Moussa C, et al. Congenital prepubic sinus. Eur J Pediatr Surg 1994;4:119-21.
9. Chou TD, Chu CC, Diau GY, et al. Subpubic sinus: a remnant of cloaca. J Urol 1995;153:1671-2.
10. Walther MM, Woodard JR. Subpubic fistula: a urethral duplication. J Urol 1996;155:1728-9.
11. Green JS, Maddon NP. Congenital prepubic sinus: a form of dorsal duplication? Br J Urol 1997;80:964.
12. Ergun O, Sayan A, Arikian A. Congenital prepubic sinus: possible variant of dorsal urethral duplication. Eur J Pediatr Surg 1998;8:380-1.
13. Nirasawa Y, Ito Y, Tanaka H, et al. Urachal cyst associated with a suprapubic sinus. Pediatr Surg Int 1999;15:275-6.
14. Huang CC, Wu WH, Chai CY, et al. Congenital prepubic sinus: a variant of dorsal urethral duplication suggested by immunohistochemical analysis. J Urol 2001;166:1876-9.
15. Soares-Oliveira M, Julia V, Aparicio LG, et al. Congenital prepubic sinus: a variant of dorsal urethral duplication (Stephens type 3). Int J Urol 2005;12:231-3.
16. Al-Wattar KM. Congenital prepubic sinus: an epispidiac variant of urethral duplication: case report and review of literature. J Pediatr Surg 2003;38:E10-2.
17. Yamada K, Kanamori Y, Tanaka H, et al. Congenital prepubic sinus: is it a residual cloacal membrane and umbilicophallic groove? Pediatr Surg Int 2004;20:47-50.
18. Usami M, Hayashi Y, Kojima Y, et al. Congenital prepubic sinus: a variant of dorsal urethral duplication (Stephens type 3). Int J Urol 2005;12:231-3.