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Congenital prepubic sinus: A case report and review of the literature

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ABSTRACT

Congenital prepubic sinus (CPS) is an extremely rare anomaly, which is often associated with purulent discharge from a midline opening overlying the pubis. CPS was first described by Campbell et al. in 1987 and they suggested that it might represent a variation in normal embryological development. Several theories have been proposed regarding the pathogenesis of CPS. However, the etiology of CPS is still unclear because the anatomical and pathological features of CPS often differ from each other. We report a case of CPS and review the literature to improve the global understanding of CPS.

1. Case report

A female neonate had a small midline opening overlying the pubis since birth. Purulent discharge was observed 1 month after birth and it then improved. After 2 years and 6 months, she was referred to our department for recurrence of discharge. She had no other symptoms and no evidence of urinary tract infection. A local examination showed a prepubic sinus with brown discharge and reddening around the sinus (Fig. 1). An ultrasound (US) examination revealed a subcutaneous cyst in front of the pubis that measured 15 × 13 × 11 mm (Fig. 2). Magnetic resonance imaging (MRI) showed a cystic lesion and thin tract to the deeper side in the subcutaneous layer (Fig. 3). We could not insert a plastic tube far enough into the sinus tract for conventional fistulography. Three months later, after the inflammation was relieved, we performed an operation. The opening of the skin was connected to the subcutaneous cyst. The sinus tract attached to the cyst extended to the pubis (Fig. 4). We injected indigo carmine into the sinus to confirm that the sinus was obstructed in front of the pubis and the sinus was excised. Histologically, the distal lumen of the sinus was lined with stratified squamous epithelium and the proximal lumen was lined with transitional epithelium (Fig. 5). The postoperative course has been uneventful without recurrence.

2. Discussion

To the best of our knowledge, there are 29 reports of CPS in the English literature (44 cases; Table 1) including 23 males and 21 females, aged 1 month to 22 years old [1–30]. CPS most commonly presents in infancy with discharge from a midline opening between the dorsal penile root/clitoris to the suprapubic region. Sinus tracts

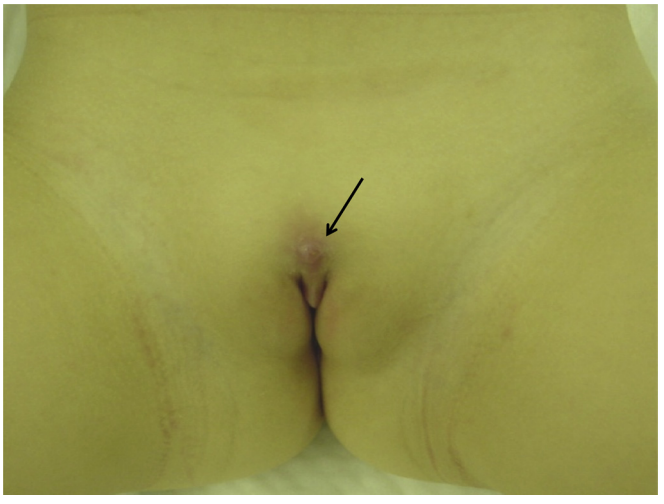

Fig. 1. Midline prepubic opening with a reddish appearance and discharge (black arrow).

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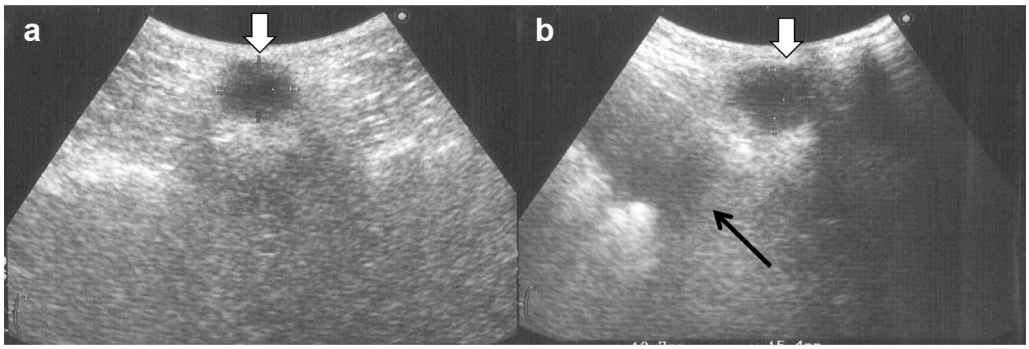

Fig. 2. US examination. (a) Horizontal scan. A subcutaneous cyst (white arrow) can be seen. (b) Sagittal scan. A subcutaneous cyst (white arrow) and bladder (black arrow) can be seen.

passed above the pubis in 16 cases, through the pubic symphysis in five cases, and below the pubis in six cases. The distal end of the sinus tract reached to the bladder wall in 25 cases, the umbilicus in five cases, the urethra in two cases, the retropubic space in five cases, the pubic symphysis in two cases, and the prepubic space in four cases. Histologically, only squamous or stratified squamous epithelium was reported in 15 cases, only transitional epithelium in six cases, both squamous/stratified squamous and transitional/urothelial epithelium in 20 cases, and cylindrical or columnar epithelium in four cases. Smooth muscle bundles surrounded the sinus in 18 cases. Connection of the sinus with the bladder was reported in two cases.

Several embryological theories have been proposed for the development of CPS. The first theory considers a localized failure of midline fusion in the lower abdominal wall [1]. During the 4th gestational week, closure of the anterior abdominal wall is complete. The cephalic border of the cloacal membrane, moving downward from the base of the umbilical cord, results in formation of the infra-umbilical anterior abdominal wall by fusion of the lateral folds. Any closure defect will cause anomalies, including omphalocele, bladder exstrophy, and epispadias. At the 9th week, a persistent cloacal membrane above the genital tubercle interrupts complete fusion, causing various anomalies, including diastasis of the pubic symphysis, bifid clitoris, separation of the corpus cavernosum, and epispadias [31]. Some cases of CPS were accompanied by diastasis of the pubic symphysis [1,18]. The presence of stratified squamous epithelium in the entire tract might support this theory.

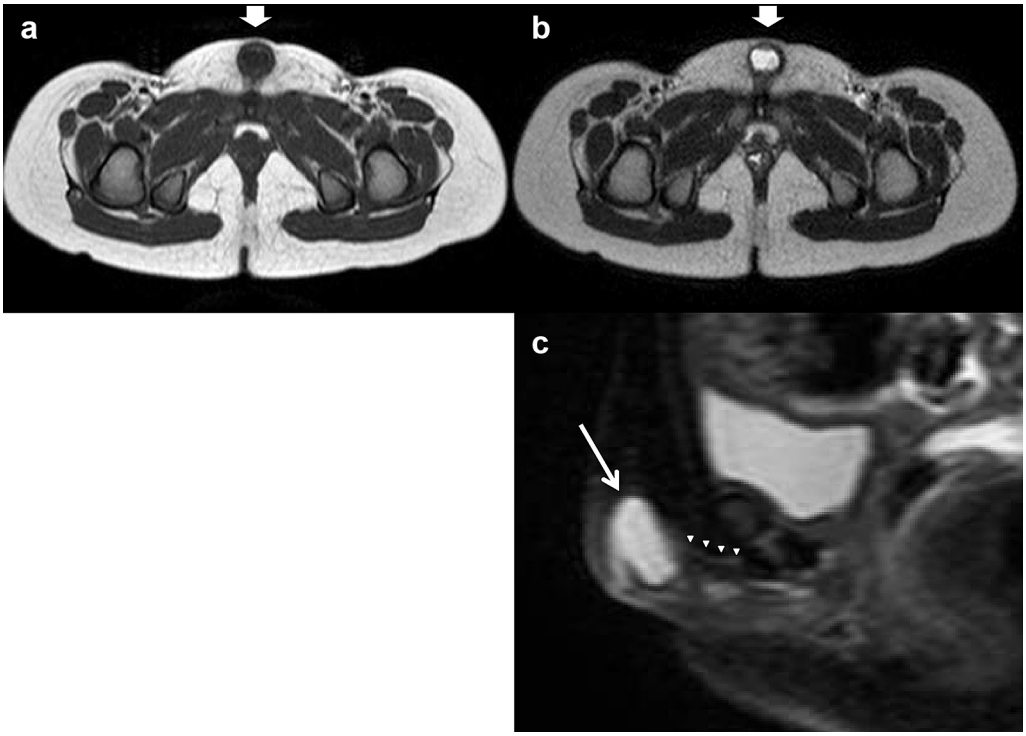

Fig. 3. MRI. (a) Horizontal scan of T1-weighted imaging. A subcutaneous cyst (white arrow) is shown. (b) Horizontal scan of T2-weighted imaging. A subcutaneous cyst (white arrow) can be seen. (c) Sagittal scan of T2-weighted imaging. A subcutaneous cyst (white arrow) and thin tract in the subcutaneous layer (white triangle) can be seen.