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Congenital Prepubic Sinus: A Case Report

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Abstract: Congenital prepubic sinus is an extremely rare congenital disorder with uncertain etiology. Also anatomic and pathologic features often differ from each other and hence classification has not been achieved. We report a case of 3 years old girl with congenital prepubic sinus which had histology of 'a dermoid sinus'.

Keywords: Congenital prepubic sinus, magnetic resonance imaging, dermoid sinus, variant of dorsal duplication of urethra.

INTRODUCTION

Congenital Prepubic Sinus was suggested as a variation in normal embryological development. It is often associated with purulent discharge from a midline opening overlying the pubis. Regarding pathogenesis of Congenital Prepubic Sinus several theories have been proposed but the etiology is still unclear because of differences in anatomical and pathological features. Till date there are only 44 reported cases in english literature. Hence we report on a 3-year-old girl with congenital prepubic sinus because of it's rarity.

CASE HISTORY

A 3 years old girl presented to our outpatient department with a small midline opening overlying the pubis with intermittent discharge from it. The complaints were since birth for which medical advice was not taken any time. She had no symptoms suggestive of urinary tract infection. Local examination revealed a prepubic sinus with white, non-foul smelling discharge (Fig. 1). Cannulation of the tract was not possible beyond 0.3 cm and hence sinogram was not performed. An ultrasound examination revealed a subcutaneous hypo echoic tract of 1.4 cm length and 1.6 mm width. The ultrasound was done twice among which the first revealed a small collection of 3*7 mm in prevescical space and second, which was done 3 weeks after first ultrasound, showed it's complete resolution. Magnetic resonance imaging (MRI) showed a sinus tract of maximum diameter of 3 mm in hypogastrium in midline and in subcutaneous plane reaching up to pubic symphysis (Fig. 2). CT sinogram revealed filling of sinus tract up to length of 0.5cm with non-opacification of the rest of the tract and with no intra-abdominal extension. Underlying bones were found normal.

Surgical exploration was performed through a circumlesional elliptical incision and after injecting methylene blue dye through external opening. A 3 cm-long sinus apparently ending as a fibrous tract at the anterior surface of the pubic symphysis was found and resected (Fig. 3). Histologically the sinus was having both keratinising stratified squamous epithelium and transitional epithelium (Fig. 4) and hence it proved to be a 'dermoid sinus' presenting as 'a variant of dorsal duplication of urethra'.

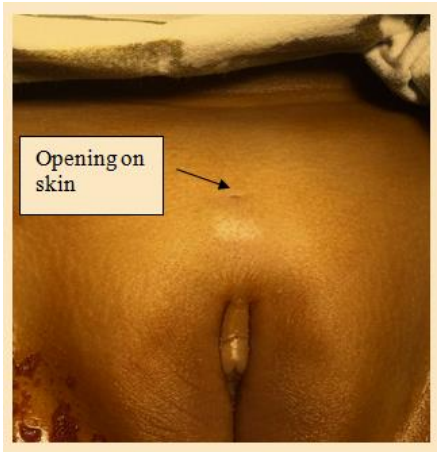

Fig-1

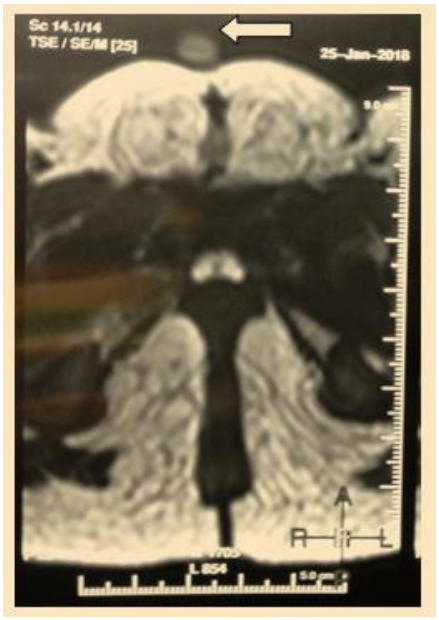

Fig-2
Arrow showing radio-opaque marker

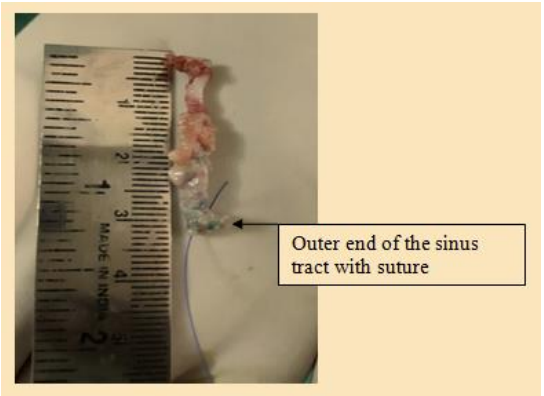

Fig-3

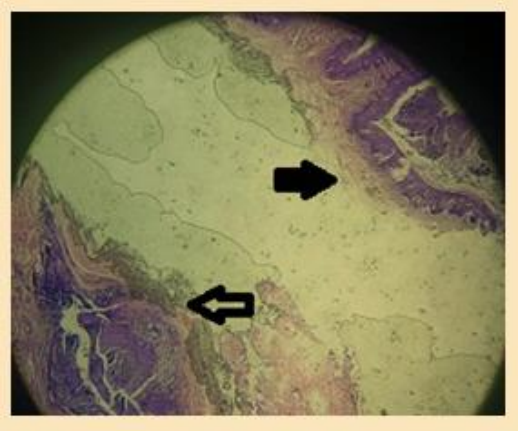

Fig-4
(Solid arrow- squamous epithelium
Hollow arrow- transitional epithelium)

DISCUSSION

Campbell *et al*. [1] was the first to describe CPS (congenital Prepubic Sinus) in 1987 and it was suggested as a variation in normal embryological development.

The development of CPS has been proposed by various embryological theories.

- Localized failure of midline fusion in the lower abdominal wall [1]: A persistent cloacal membrane above the genital tubercle, at the 9th week of gestation, interrupts complete fusion, causing various anomalies such as diastasis of the pubic symphysis [1, 2], bifid clitoris, separation of the corpus cavernosum, and epispadias [3]. Presence of stratified squamous epithelium in entire tract supports this theory.
- A variant of dorsal duplication of urethra [1]: Stephens [3] described three types of dorsal urethral duplication according to the anatomy. Type 1- a complete or incomplete tandem channel that runs parallel to normal urethra from glans to bladder and joins urethra or ends blindly. Type 2- an epispadiac type of channel [2, 5] from the dorsum of penis reaching up to bladder or it joins urethra at some point. Type 3- a dermoid sinus [2, 5] that simulates accessory urethra, but tracks from base of penis in front of pelvic urethra and bladder, behind the pubic symphysis, toward or up to umbilicus. Presence of transitional urothelium in the proximal part of the sinus favours this theory [2].
- A congenital fistula of primitive urogenital sinus as described by Soares-Oliveira *et al.* [4]
- A residual cloacal membrane in umbilico-phallic groove [2].

CPS most commonly presents in infancy as a midline opening, between the dorsal penile root or clitoris and suprapubic region, with discharge. Till today there are only few cases reported.

Anatomically, sinus tract either passes above the pubis; through the pubic symphysis or below the pubis. Also the distal end of sinus tract reaches either to the bladder wall or to retropubic space or to pubic symphysis or to prepubic space or to urethra or to umbilicus.

Histologically, it shows either only squamous/ stratified squamous epithelium or only transitional epithelium or both squamous/stratified squamous and transitional or cylindrical/ columnar epithelium. Sometimes smooth muscle bundles surround the sinus tract.

Magnetic Resonance Imaging (MRI) is the diagnosis of choice for obtaining all anatomical details of CPS [2]. Complete excision is the curative treatment.

Complications, if the tract left unexcised, are infection [2] presenting as prevescical abscess, late malignant changes [5] (as per epithelial origin).

In our case, the sinus tract reached up to pubic symphysis. Histology of the excised tract showed both keratinising stratified squamous epithelium and transitional epithelium.

CONCLUSION

CPS is rare congenital entity for which complete excision is simple and the curative treatment, to prevent recurrence of symptoms and possible complications.

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