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Case report

Congenital prepubic sinus – A variant of urethral duplication

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Abstract
Introduction: Congenital prepubic sinus (CPS) is a rare anomaly. It has been considered as one of the presentations of the spectrum of vesico urethral developmental defects.
Observations: We are reporting two such rare cases: one in a year old female and another in a nine and half year's old male child.
Conclusion: Simple excision of the sinus tract is preferred in most patients. Awareness and knowledge of the anatomical variations of the course of the sinus tract will help in complete excision and hence avoiding the recurrence.

Introduction

Congenital prepubic sinus (CPS) is a rare condition. It has been referred in the literature with various names like, congenital prepubic sinus, subpubic fistula and prepubic dermoid sinus. The aetiopathogenesis is still not clear, but it has been considered as one of the variations of the dorsal urethral duplication. They usually present with discharge from the sinus. Excision of the sinus tract is the treatment of choice.

Case series

Case 1

One year female baby was brought with history of intermittent discharge from an opening in the pubic area since neonatal period. Baby was born normally at term and mother did notice a tiny opening at the pubic area within few days after birth, without having any voiding disturbances. Clinical examination revealed small opening in the prepubic are, around 1 cm above clitoris without any

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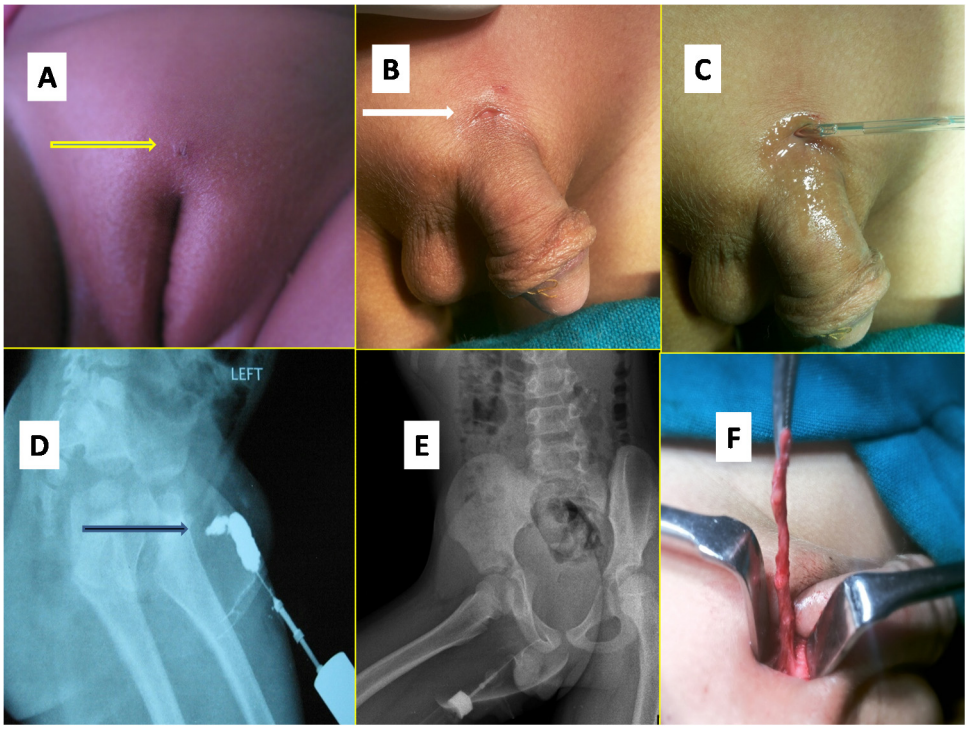

Figure 1 A- Yellow colored arrow indicating the sinus opening in a female child. B-White colored arrow indicating sinus opening in male child. C-Tip of the catheter in the sinus opening. D-Black colored arrow indicating the blind ending sinus tract. E-Sinogram in male child; revealing non communication to the urinary tract. F-Intra-operative picture showing the sinus tract.

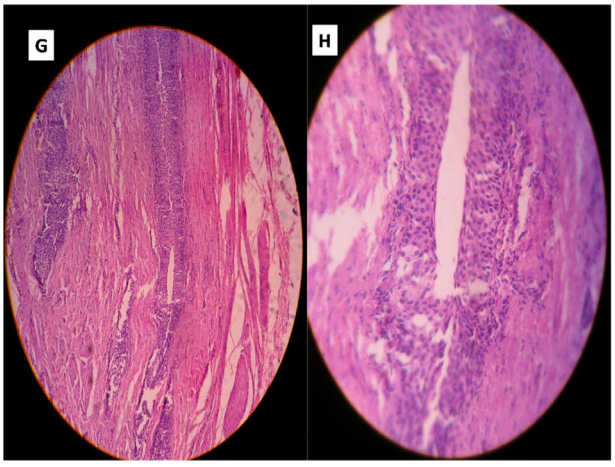

Figure 2 Histopathological picture. G-low power field showing transitional urothelium. H-high power field showing the transitional urothelium.

discharge (Fig. 1A). The external genitalia, perineum and the spine were normal. Screening ultrasonography of abdomen and pelvis was also normal. Sinogram delineated a short sinus tract ending with a bulbous dilatation; there was no communication with urethra or bladder (Fig. 1D). Baby underwent excision of the sinus tract and had uneventful recovery. Histology of the tract showed urothelium with smooth muscle bundles (Fig. 2G). Baby was doing well at 5 years follow up.

Case 2

A 9 ½ yr boy was brought with complaint of an abnormal opening in the pubic area noticed since early infancy with history of intermittent clear discharge. On examination, the external genitalia was normal with circumcised penis. A tiny opening was seen over the prepubic area 1 cm from the base of the penis (Fig. 1B). Renal ultrasound and Micturating cysto urethrography were essentially normal. Sinogram revealed the blind ending tract measuring around 1.5 cm without any connections to urethra or bladder (Fig. 1E). Intra-operatively the sinus tract was measuring 4 cm in length when stretched, traversing the rectus sheath and was ending blindly anterior to bladder (Fig. 1F). The tract was excised completely and baby had uneventful recovery. Histology revealed urothelium surrounded by smooth muscle bundles with minimal inflammation (Fig. 2H). Patient was doing well at 7 months follow up.

Discussion

Congenital prepubic sinus is a rare congenital anomaly. Only 39 cases have been reported in the English literature since it was first described by Campbell et al in 1987 [1,2]. Only 17 cases have been reported in females till 2013 [2]. Embryo-pathogenesis of CPS is still unclear though numerous theories have been postulated in the literature, which include; 1. Anomaly of the abdominal wall closure, 2. Developmental defect of the urethra, similar to duplication of urethra and 3. Fistula of the primitive urogenital sinus, which has three anatomic subtypes depending on the direction of the sinus tract, a. high; towards the urachal remnant, b. middle; towards the bladder and c. low; towards the prostatic urethra [1,4]. Tsukamoto et al in 2004 has postulated that CPS may be caused by a residual

cloacal membrane and umbilicophallic groove [1,5]. Huang et al reinforced the theory of dorsal urethral duplication by immunohistochemical staining technique, which was done on the excised sinus tracts [6]. They found transitional epithelium in the proximal part of the sinus with surrounding smooth muscle bundles [5]. However, Campbell et al reported three cases of CPS in which the sinus tract was surrounded by stratified squamous epithelium or transitional epithelium with concentric bundles of collagen and smooth muscle fibers [3]. Stephens described 3 types of dorsal urethral duplications according to their anatomy. Type 1 is a complete or incomplete tandem channel, which runs parallel to the normal urethra from the glans to the bladder, joins the urethra or may end blindly. Type 2 is an epispadiac type of channel from the dorsum of the penis to the bladder or one that joins the urethra at some point. Type 3 is a dermoid sinus that simulates an accessory urethra but tracks from the base of the penis in front of the pelvic urethra and bladder, behind the pubic symphysis to or towards the umbilicus [2,7]. Of all the theories, most authors favor the theory of dorsal urethral duplications [1–3].

The operative findings in both of our cases were similar to type 3 of Stephens's classification. In both cases, the sinus tract was traversing towards the undersurface of the pubic symphysis through the rectus sheath stopping just anterior to bladder without any communication. In both of our cases, histology revealed transitional epithelium with few smooth muscle bundles pointing towards the urethral origin.

Diagnosis is mainly clinical, one should consider the possibility of CPS in all patients with sinus in the prepubic area and definitive diagnosis requires confirmation by histology. The imaging techniques such as Sinogram and Voiding cystourethrogram will be helpful in cases with long sinus tract with retropubic extension, communication with urinary tract or rarely intra-abdominal extension if any [2]. Routine use of sinogram may be useful as it is difficult to differentiate between simple tracts, from the ones with deeper extension only on clinical basis [2]. In both of our cases the imaging did not reveal any communication with the urinary tract; the tract in the first case had larger diameter compared to the second one (Fig. 1D, E).

Treatment of choice is excision of the sinus tract and is curative [1–5]. In most case the surgical technique is simple, but in cases with retropubic extension, one has to consider combined perineal or intra-abdominal approach, if required [2].

Conclusion

Congenital prepubic sinus is an unusual congenital anomaly. It is considered as a variant of dorsal urethral duplication. Excision of the tract is treatment of choice; which is simple and curative. Awareness and knowledge of the anatomical variations of the course of the sinus tract will help in complete excision and hence avoiding the recurrence.

The ethical committee approval

This study obtained the approval of the ethical committee.

Authors' contribution

Dr J. Aihole: operating on the patient, the concept, collection of data, collecting the literature, counseling the patients attenders, writing the article, preparing the article and submitting the article.

Dr N. Munianjanappa: operating on the patient, critically reviewing the article and grammar correction.

Dr D. Javaregowda: critically reviewing the article, grammar correction.

Dr V. Jadhav: critically reviewing the article, helping to submit the article.

Conflict of interest

None.

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