



Congenital prepubic sinus: Report of two cases

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ABSTRACT

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Congenital prepubic sinus is a rare disorder with fewer than 30 cases reported in the literature. The origin of the sinus and its tract is unclear but urethral duplication, cloacal remnant or midline closure defect are amongst the postulated mechanisms for the development of the sinus. Chronic discharge and infection are the main indications for surgery. Although surgery is the definite way of correction, timing and extent of surgery are not well documented due to the limited number of cases. Here we present two cases with congenital prepubic sinus, a four month old girl and a five years old boy, who were admitted with discharge through the external opening of the prepubic sinus which are shown not to have a connection with urinary system and are managed by limited surgical excision. Postoperative courses were uneventful and no relapse or complications were encountered. Histological examination revealed that the sinus tract is lined with squamous stratified epithelium without any other significant finding. Although the classification and definition of congenital prepubic sinus is not clear, extensive imaging modalities and surgical procedures are not necessary in most conditions as these can create additional burden for the patient and the healthcare system and may cause complications at the cost of nothing.

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1. Introduction

Congenital prepubic sinus is a rare disorder with fewer than 30 cases reported. It is usually diagnosed during infancy. The most common presenting symptoms are discharge from the sinus and recognition of the sinus orifice by the parents. Although no malignancy is reported related with prepubic sinus, chronic discharge, infection of the sinus and even abscess development may be annoying (Kobayashi et al., 2015; Shaw et al., 2015). Chronic discharge and infection are the main indications for surgery. Here we present a four month old girl and a five year old boy, both were admitted with

intermittent whitish discharge from the prepubic sinus orifice.

2. Case report

Patient 1 was an otherwise healthy four month old girl with no history of urinary tract infection. Urine examination and ultrasonographic investigation of the kidneys and bladder were normal. On physical examination, external genitalia was normal with a tiny skin dimple on the midline about 1 cm cranial to the anterior commissure of major labia without an obvious opening (Fig. 1). Diagnostic cystoscopy was normal.

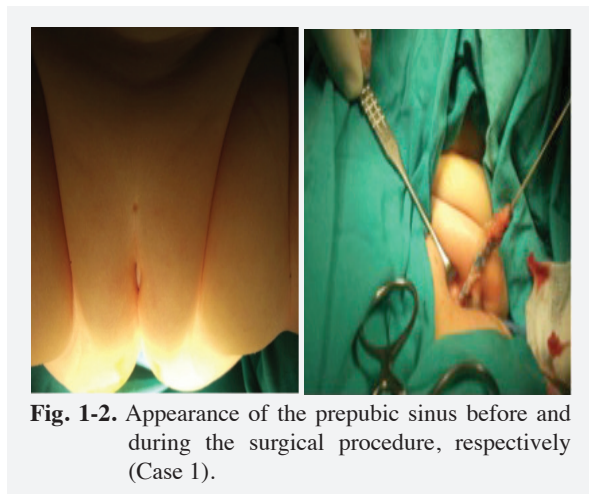


Fig. 1-2. Appearance of the prepubic sinus before and during the surgical procedure, respectively (Case 1).

Simultaneously the dimple was cannulated with a 24G venous catheter and methylene blue was injected but no connection could be demonstrated between the sinus and bladder or urethra. After surgical preparation, sinus tract was explored by a circumferential incision. Sinus tract was about 3 cm long and blindly ending by a fibrous band under the pubic symphysis where the exploration was completed and tract was excised (Fig. 2). Layers were closed appropriately without leaving dead space behind. The postoperative course was uneventful.

On histological examination, sinus tract was lined with squamous stratified epithelium with mononuclear cell infiltration but no surrounding muscle structure was observed.

Patient 2 was an otherwise healthy five years old boy admitted with discharge through the sinus and without any history of urinary tract infection and is fully continent (Fig. 3). He underwent a voiding cystourethrography examination which was totally normal. Diagnostic cystoscopy is performed and no connection is demonstrated with the urinary tract by methylene blue injection through the sinus orifice. On surgery, sinus tract is excised by a circumferential incision which was 2.5 cm long (Fig. 4). The postoperative course was uneventful.

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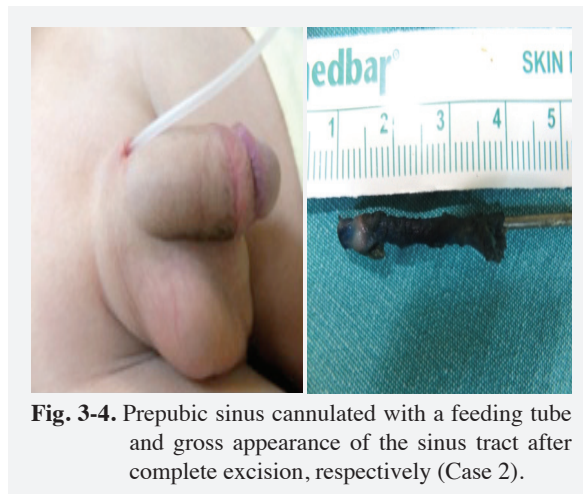


Fig. 3-4. Prepubic sinus cannulated with a feeding tube and gross appearance of the sinus tract after complete excision, respectively (Case 2).

Histological examination revealed that sinus tract was lined with squamous stratified.

3. Discussion

The origin of the prepubic sinus and its tract is unclear but urethral duplication, cloacal remnant or midline closure defect are amongst the postulated mechanisms for the development of the sinus (Rozanski et al., 1990; Chao et al., 2002; Soares-Oliveira et al., 2002; Shaw et al., 2015). These theories depend on the histological findings of the sinus but must be interpreted cautiously as epithelium may change depending to the exposed environment. Urethral duplication theory is also not satisfactory as proximal part of the tract is blindly ending and fibrotic in most of the cases (Tander et al., 1993; Shaw et al., 2015).

Although the origin of prepubic sinus is unclear, simple surgical excision avoiding an extended surgical procedure is recommended. Overactive bladder syndrome development after the surgery is reported (Soares-Oliveira et al., 2002; Shaw et al., 2015). As in our series, limited surgical exploration is sufficient and effective without compromising success. Nevertheless, complicated imaging modalities have limited contribution as it is not connected to the urinary system or other organ systems.