Case Report
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Congenital Prepubic Sinus: A Case Report

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Summary

Congenital prepubic sinus is an exceptional disorder of uncertain origin. These sinuses may represent a variant of epispadiac duplication or dorsal urethral duplication of the urethra. We present a case of isolated blind ending prepubic congenital sinus in a 5-year-old male child, which revealed transitional epithelium lining proximally, and squamous epithelium distally. This entity presents with either an opening in the prepubic region and/or persistent mucoid discharge. Excision leads to a permanent cure.

Key Words: Congenital prepubic sinus, Urethral duplication, Epispadias.

Introduction

Congenital prepubic sinus is a rare disorder of uncertain etiology. They are usually lined by transitional and squamous epithelium[1]. There have been few cases in the literature since the first report by Campbell et al in 1987[2]. These sinuses present as confusing small tract, which commences in the skin overlying pubic symphysis, and extends directly to the anterior wall of the bladder[1] or the umbilicus[3]. We present a case of isolated blind ending prepubic congenital sinus extending towards the bladder.

Case report

A 5-year old boy presented with a congenital sinus over the dorsal aspect of the base of the penis. There was history of occasional mucoid discharge from the sinus opening. There was no history of urinary tract infection. A sinogram revealed a 4 cm tract extending towards the anterior wall of the urinary bladder without any communication with bladder or urethra. A micturating cystourethrogram showed a normal bladder and urethra. An infant feeding tube could be inserted 4 cm up the sinus tract (Fig 1). Complete excision of the tract was done. The 4 cm long track ended blindly below the pubic symphysis. Histological examination of the specimen showed a tract lined with both transitional epithelium (proximal portion) and squamous epithelium (distal portion) with a thin layer of surrounding smooth muscle.



Fig 1: Clinical photograph showing cannulated prepubic sinus

Discussion

Congenital prepubic sinus is a rare anomaly. Embryologically these sinuses may represent a variant

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of epispadiac anomaly[2] or dorsal urethral duplication[3]. Depending upon the anatomic and embryologic features, Stephens classified the dorsal urethral duplications into three types[4]. Type 1 is a complete or incomplete channel running close or parallel to the normal urethra from the glans to the bladder. Type 2 is described as an epispadiac channel that joins either the bladder or the normal urethra. Type 3 is the most unusual anomaly, in which a dermoid sinus that simulates a urethra but tracks from the base of the penis or clitoris anterior to the pelvis, urethra and bladder, behind the pubic symphysis to or towards the umbilicus. Our case represents a type 3 anomaly. However Tsukamoto K et al postulated that congenital prepubic sinus may be caused by a residual cloacal membrane and umbilicophallic groove and that its depth may determine the position of its ending[5]. Using the immunohistochemical staining technique of the excised sinus Huang et al found presence of transitional epithelium in the proximal part of the sinus with smooth muscle bundles around it reinforcing the theory that congenital prepubic sinus is a variant form of dorsal urethral duplication only1. Treatment consists of total excision of the accessory urethra by way of suprapubic approach without symphysiotomy as done in our case[6]. The surgical

Le sinus prépubien congénital: un rapport de cas

Résumé

Le sinus prépubien congénital est un désordre exceptionnel d'origine incertaine. Ces sinus peuvent représenter une variante de la reproduction de l'épispadias ou de l'urètre dorsal. Nous présentons un cas de sinus prépubien congénital isolé sans issue chez un garçonnet de 5 ans, qui a révélé proximalement une paroi d'épithélium transitionnel, et distalement une paroi d'épithélium squameux. Cette entité se manifeste soit par une ouverture dans la région prépubienne et/ou par un écoulement muqueux persistant. L'excision mène à une guérison permanente. ***Mots-clés:*** Sinus prépubien congénital, reproduction urétrale, épispadias.

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