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PEDIATRIC UROLOGY
Case Report

Congenital prepubic urachal-cutaneous fistula

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ABSTRACT

Congenital prepubic fistula is a very rare congenital anomaly that manifests as a punctiform opening in the midline of the pubic region. According to Campbell, congenital prepubic fistulae are duplicates of the dorsal urethra. Recent investigations have shown that this problem may have a complex etiology. We report a case of a congenital prepubic fistula in 2-month old female neonate, which manifested as a simple punctiform opening in the pubic region, with purulent secretion that resulted from applying pressure to the region. The surgical treatment was successful, and the infant had an uneventful postoperative course of healing.

Keywords: Congenital; prepubic fistula; treatment.

Introduction

Congenital prepubic fistula is a rare congenital anomaly that manifests as a punctiform opening in the midline of the pubic region. Only 40 cases of this congenital anomaly have been reported from 1970 to 2015, according to the PubMed database.[1-14] The first description of this anomaly was given by Campbell, who regarded it as a duplicate of the dorsal urethra.[2] Recent investigations have shown that this problem may have a complex etiology.[3,4,15] We report a case of a congenital prepubic fistula in 2-month old female neonate, which manifested as a simple punctiform opening in the pubic region, with purulent secretion that resulted from applying pressure to the region.

Case presentation

A 2-month old female baby was referred to our center for the investigation and treatment of a punctiform opening in her pubic region (Figure 1a). Application of pressure to the pubic region resulted in a purulent secretion from the opening (Figure 1b). Diagnostic procedures included laboratory investigations, ultrasonography (US) of the anterior abdominal wall and abdomen, voiding cystourethrogram (VCUG), fistulography, and cystoscopy. Fistulography using a hydrosoluble contrast showed a cutaneous fistula with urachal communication (Figure 1c). This tract connected the fistula to the distal/caudal part of the urachal remnant of about 6 cm length and 5 mm width, without any other connections with other tissues. The US, VCUG, and cystoscopy did not reveal any pathological findings. The surgical procedure was started with an injection of methylene blue dye into the sinus via the punctiform opening in the skin, followed by a 3 cm long upper abdominal transverse incision. The sinus tract was identified inside the subcutaneous tissue. An additional small fusiform incision was made around the punctiform opening and the sinus was dissected, pulled out, and completely removed through the primary incision (Figure 2). The tract was connected to the rectus sheath at the midline and length-wise to the median umbilical ligament (its caudal segment), which made it seem similar to the urachal remnant. Histopathological examination revealed that the lesion was lined by the transitional epithelium, therefore, we diagnosed the urachal remnant. The infant had an uneventful postoperative course of healing.

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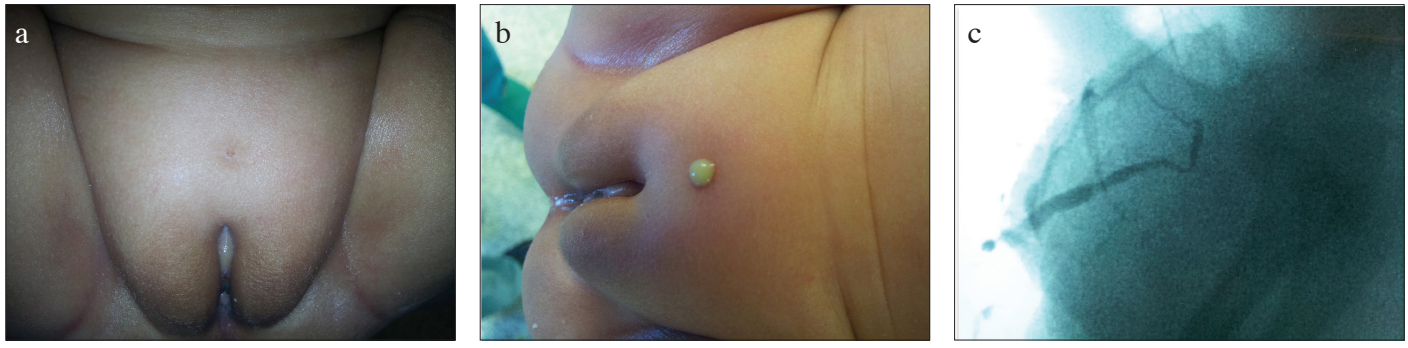

Figure 1. a-c. Preoperative view of a punctiform opening in the pubic region (a), the purulent secretion from the opening (b), and fistulography (c)

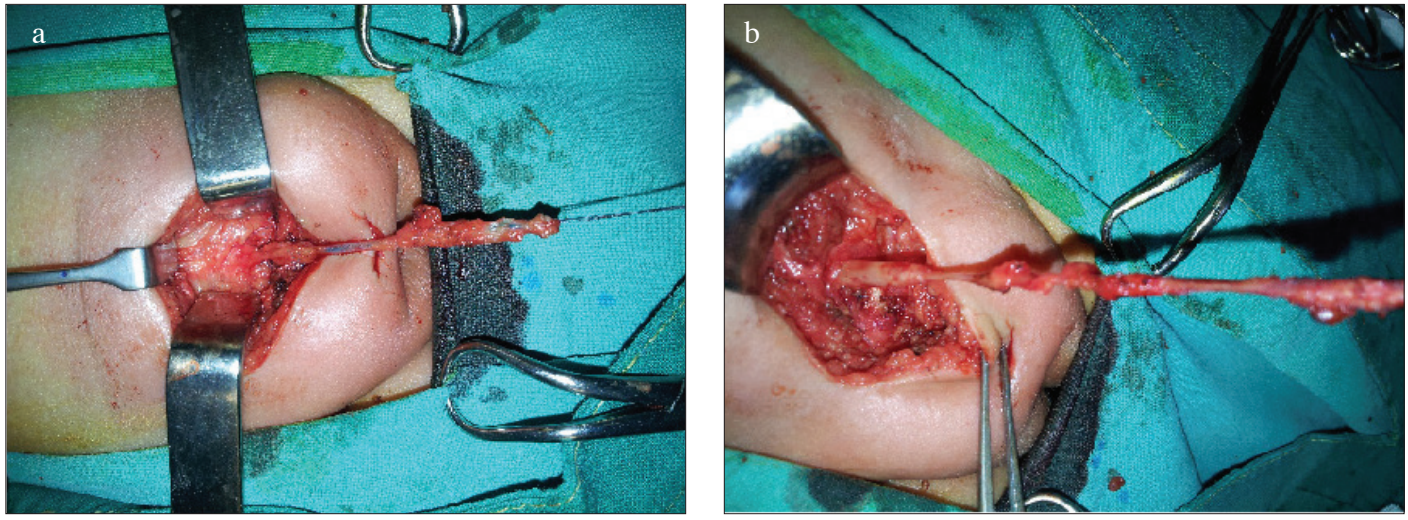

Figure 2. a, b. Complete excision of the fistula

Discussion

There are four generally accepted theories for the congenital prepubic urachal-cutaneous fistula (CPUCF) etiology. The first theory suggests that CPUCF is a result of the incomplete abdominal wall fusion below the umbilicus during embryogenesis.[5,6] The second theory proposes that CPUCF is a urethral developmental anomaly (a variant of the dorsal urethral duplication).[1,2,7-10] The third theory states that CPUCF is a congenital fistula of the primitive urogenital sinus, which may present as three anatomical subtypes: 1) low-type: toward the urethra; 2) middle-type: toward the urinary bladder, and 3) high-type: toward the urachal remnant.[3,11,12] The fourth theory suggests that CPUCF occurs as a result of the incomplete involution of the cloaca and its remnants.[4,13,14] All of these theories have their own functional and embryological proofs, but neither one of them gives a clear explanation for the occurrence of this anomaly. Only 40 cases of CPUCF have been reported so far in the scientific literature (male: 22, female: 18).

In conclusion, based on our study, we conclude that this anomaly is a result of incomplete obliteration of the primitive urogenital sinus and manifests the high-type fistula, i.e., it is present towards the urachal remnant, which supports the third developmental theory of this anomaly. Generally speaking, all of these theories have their own justified and unjustified proofs, but there are not enough reported cases to perform a detailed analysis. Therefore, all patients with this anomaly should be presented in order to achieve a sufficient number of cases for a more detailed analysis in the future.

Informed Consent: Written informed consent was obtained from the parents of the patient whose case was reported.

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