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Urethral Caruncle with Associated Renoureteric Anomalies

Sohail Ahmad, *Akash B. Pati, Santosh K. Mahalik, Kanishka Das

Department of Pediatric Surgery, All India Institute of Medical Sciences, Bhubaneswar, India
**Corresponding Author's e-mail: patiakash.ap@gmail.com*

Abstract

A urethral caruncle is a rare differential diagnosis for a prolapsed mass at the urethral meatus in a prepubertal girl. The aetiopathogenesis of this entity is unclear. We describe the presentation and treatment of a patient with a urethral caruncle and associated renal anomalies that have not been described earlier. The aetiology and treatment of the entity has also been discussed.

Keywords: Urethral Caruncle, Premature menarche, Paediatric Bleeding per vagina, Crossed fused ectopia

Introduction

Prepubertal bleeding at the introitus is infrequent and raises concern among parents and health providers alike. A wide variety of differential diagnoses are considered from an innocuous abrasion to malignancy. Urethral etiologies include mucosal prolapse or polyps. A caruncle isa rare cause that is commoner in middle aged or postmenopausal women. Of 14 cases of urethral caruncle in pediatric age group described in English literature, only 4 have been documented [1] in detail, and none had an associated upper urinary tract anomaly. We describe a patient with a urethral caruncle, bleeding per urethra and associated upper urinary tract anomaly.

Case Report

A 3–year female with normal perinatal and past medical history presented with intermittent

bleeding from the introitus for a month. There was no history of sexual abuse, genital trauma or recurrent urinary tract infections. In the interim, she was asymptomatic. General physical examination and abdominal examination was unremarkable. The external genitalia were phenotypically female; however, the urethral meatus was circumferentially large and exuberant. The mucosa was relatively more everted along the inferior meatal margin. (Figure 1- A, B).

The results of routine hematological and biochemical examinations were normal. The urinalysis showed plenty of red blood cells. An ultrasonogram revealed a small left kidney (45X20mm), a larger right kidney (69X28mm) and crossed fused renal ectopia. Renal cortical scintigraphy (dimercapto succinic acid) showed a left to right crossed fused renal ectopia (Figure -2). The left renal unit was hydronephrotic and had mildly impaired cortical function and a mid-polar cortical irregularity. The differential function was 43% in the left renal unit and 57% in the right renal unit which was normal. A voiding cystourethrogram was normal.

Examination under anesthesia and cystourethroscopy was performed. The urethral meatus was wide (diameter 1.8cm) with its edges uniformly protuberant and firm. The mucosa was relatively more everted at the inferior circumference. Cystoscopy revealed anormal urethra with a mildly trabeculated bladder. The right ureteric orifice was normal, whereas the left was grossly superolateral and patulous. Vaginoscopy was unremarkable.

The patient was discharged on Sitz bath (advised to sit immersed in a warm water bath up to hip, aids in reducing congestion) twice daily and advised to apply 0.1% betamethasone creamlocally once daily. By 6 weeks of therapy, there was a significant reduction in size of the everted mucosa which appeared normal. At 2 years follow up, there has been no further grosshematuria and the lesion has disappeared completely. The initial hydronephrosis is non progressive and renal function is stable on scintigraphy. Informed consent was obtained from the parents for publication of this case report.

Discussion

The urethral caruncle is a common benign tumor of the female urethra usually seen in postmenopausal women. An occurrence in the premenarche period, as described here, is rare. The

caruncle appears like a raspberry protruding from a quadrant commonly posterior wall ofthe urethral circumference. It is rarely observed at other locations.[2] A circumferential caruncle mimicking a urethral prolapse has been reported only twice since 1964. [1]

The exact etiology of urethral caruncle is unknown; however, chronic inflammation and estrogen deficiency have been implicated.[1] According to Jeffcott, a true caruncle is a vascularpapilloma that arises as a polyp from the posterior lip of the urethra, whereas a pseudo caruncleis a granuloma arising as a diffuse, sessile red lesion. [3] Urethral caruncles have been reported at birth; hence, a congenital origin is possible. [4]

Symptoms include pain during micturition (51%), bleeding (49%), a mass at the meatus (41%)and an increase in urinary frequency and urgency (36%)[3] A 'premature menarche' without other secondary sexual characters may point towards a bleeding urethral caruncle.[5] The patientdescribed herein had gross intermittent hematuria and a prominent peri meatal mass. The clinical differential diagnosis for a periurethral mass includes urethral prolapse, prolapsing ureterocele, and a botryoid bladder or vaginal rhabdomyosarcoma. A urethral prolapse

protrudes circumferentially around the meatus like a soft rosette with a central dimple.[6] Also, both urethral prolapse and polyp are mucosa covered. In contrast, a caruncle is covered with granulation tissue and is liable to bleed. Microscopically, the urethral caruncle is a bed of granulation tissue that may feature squamous or transitional epithelium at places. Besides, marked inflammatory infiltrate and vascular engorgement of the stroma is common.

Most authors consider them as acquired anomalies. [1,7] The present case features an ectopic left kidney, ectopic termination of left ureter, and a urethral mega meatus. To the best of our knowledge, such anomalies have not been previously reported. We suggest that patients with urethral caruncle need to be evaluated for associated anomalies in the upper urinary tract. No reports available regarding malignancy in urethral caruncle in children. However, in adults, 2.4% of all patients with a preoperative diagnosis of urethral caruncle were found to have carcinoma.[8]

Various treatment modalities are advocated ranging from conservative management with Sitzbath,

estrogen creams, topical corticosteroids to surgery. [1,5,7] In unresponsive cases or those exhibiting a progressive, irregular or suspicious growth, electrocoagulation of the base or excision are surgical options. [1] Surgical excision may cause urethral stenosis in a circumferential lesion. The present case responded favorably to the initial conservative management.

Conclusion

The patient had a granulated mass at the posterior margin of the circumference of the urethral meatus that was diagnosed as a urethral caruncle. She presented with hematuria instead of the typical blood spotting. It was associated with a crossed fused renal ectopia and a routine ultrasonographic screening is suggested to detect associated reno ureteric anomaly. It was managed conservatively with steroid creams and Sitz baths.

Authors' Contributions

All authors were involved in the conceptualization and preparing of the manuscript. All authors approved the final version.

References

1. Chiba M, Toki A, Sugiyanma A, Suganuma R, Osawa S, Ishii R et al . Urethral caruncle in a 9-year-old girl: A case report and review of the literature. J med casereports, 2015: 28:9 https://doi.org/10.1186/s13256-015-0518-7
2. Turkeri L, Simsek F, Akdas A. Urethral caruncle in an unusual location occurring in prepubertal girl.Eur. Urol. 1989;16:153-4 https://doi.org/10.1159/0004715553
3. Becker LE. Urethral caruncle: a herald lesion for distal urethral stenosis? J Natl Med Assoc. 1975; 67: 228-30
4. Jarvi OH, Marin S, De Boer WG. Further studies in intestinal heterotopia in urethral caruncle. Acta Pathol Microbiol Immunol Scand.1984;92(6):469 https://doi.org/10.111/j4.tb04429.x
5. Manori G, Beneragama D. Urethral caruncle presented as premature menarche in a 4-year-old girl. Case report in pediatrics. 2018, https://doi.org/10.1155/2018/34860326
6. Murphy JP, Gatti JM. Abnormalities of the urethra, penis and scrotum. In : Coran AG editor Pediatric Surgery 7th ed Philadelphia Elseiver Saunders; 2o12. P 1558

7. Kim KK, Sin DY, Park HW. Urethral caruncle occurring in a young girl: A case report. J Korean Med Sci. 1993;8:160-1 https://doi.org/10.3346/jkms.1993.8.2.160
8. Marshall FC, Uson AC, Melicow MM. Neoplasms and caruncles of the female urethra. Surg Gynecol Obstet. 1960 ;110:723-33. PMID: 14421527.

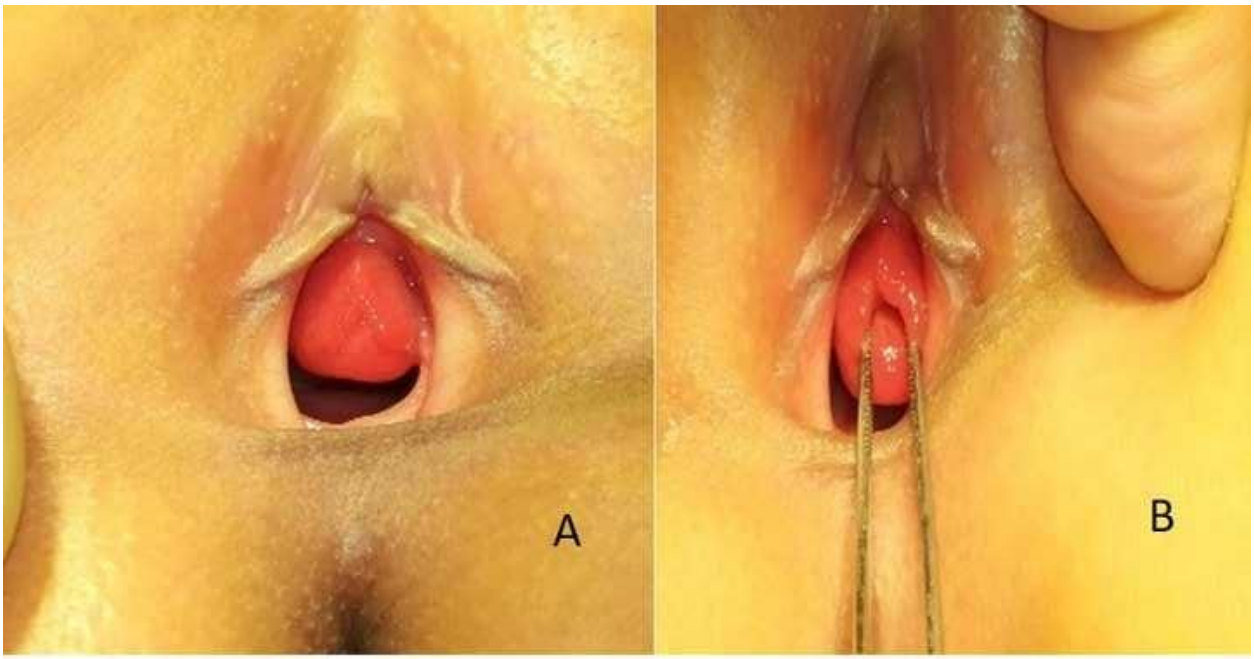

Figure 1: A: Hyperaemic exuberant mucosa at the urethral meatus. **B**: The exuberant mucosa was more prononced inferiorly

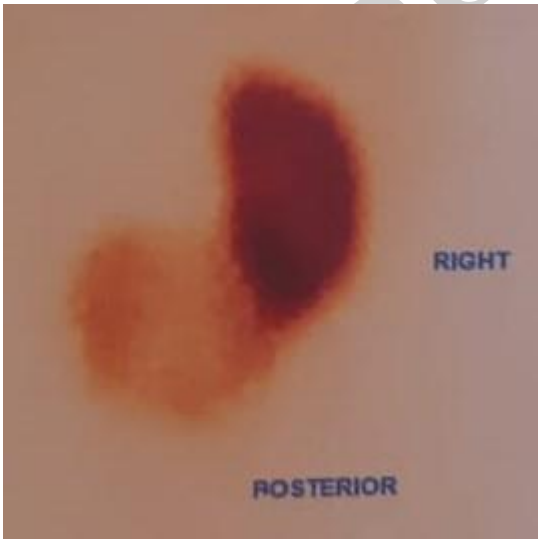

Figure 2: Renal Scintigraphy showing left to right crossed fused renal ectopia.