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Bladder outlet obstruction secondary to posterior urethral cystitis cystica & glandularis in a 12-year-old boy. A rare case scenario

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ABSTRACT

Urethral cystitis cystica and cystitis glandularis in children are extremely rare condition. Mainly manifested as weak urine flow. Other symptoms may occur, such as terminal hematuria, urgency, frequency, and urinary incontinence. In ultrasound examinations, children usually have high post-void residual urine volume. Transurethral visualization and resection of lesions is a method of diagnosis and treatment. Close follow-up is necessary because recurrence may occur. In addition, systemic corticosteroids may slow down its regrowth.

In the current case report, we discussed the clinical presentation and management of urethra cystitis cystica, and cystitis glandularis in a 12-year-old boy.

Introduction

Cystitis cystica is a disease characterized by hyperproliferation, in which the initial submucosal epithelial cells form masses termed "Brunns nests", which undergo cavitation to form fluid-filled cysts. This local immune hyperproliferative reaction is thought to be triggered by a chronic inflammatory process because it is related to recurrent urinary tract infection (UTI) caused by different organisms such as Escherichia Coli, Pseudomonas, Proteus and Chlamydia.[1] In adult autopsy studies, the incidence of cystitis cystica was 1.4%. However, the exact incidence in children is unknown.[1]

On the other hand, in cystitis glandularis, mucosal cells in the inner bladder wall transform into glandular metaplasia. This is a common finding in bladder biopsy and cystectomy, usually present in the trigone area. Bladder exstrophy, pelvic lipomatosis and recurrent UTI are also associated with cystitis glandularis.[2] Cystitis glandularis is very rare in children, with few case reports in the literature.[2]

The urinary bladder is the most common part affected by both lesions, and the most common locations are the lateral bladder walls, trigone and the bladder neck. Symptoms such as hematuria and/or irritative lower urinary tract symptoms (LUTS) may be the main symptoms of bladder cystitis cystica. In addition, ultrasound examination may reveal cystitis glandularis as a polypoid mass.[2] It is worth noting that cystitis cystica and cystitis glandularis are unlikely to present in the urethra. According to one study, cystitis cystica with squamous metaplasia of the urethra was reported in a 9-year-old boy, and histopathology did not show cystitis glandularis.[3]

To the best of our knowledge, the combined lesions of urethral cystitis cystica and cystitis glandularis in children have not been described before. In this case report, we discuss the management of this rare condition in a 12-year-old boy.

Case presentation

A 12-year-old boy visited our urology clinic. He complained of irritative LUTS (i.e. frequency and urgency) and weak urinary stream for more than 1 year. Then, he developed repeated episodes of terminal hematuria in the last month. There was no history of previous UTI, urinary stone disease, urological instrumentations, trauma, or any malignancy. The family history was unremarkable.

Upon examination, the patient's abdomen was soft and lax, without palpable masses. He was circumcised a with normal phallus and testicles. His laboratory blood tests, urinalysis and culture were normal. Ultrasonography of the urinary tract showed normal kidneys, but the urinary bladder showed a 1.2 × 0.8 cm well defined hyperechoic structure on the posterior wall close to the bladder neck (Fig. 1). There

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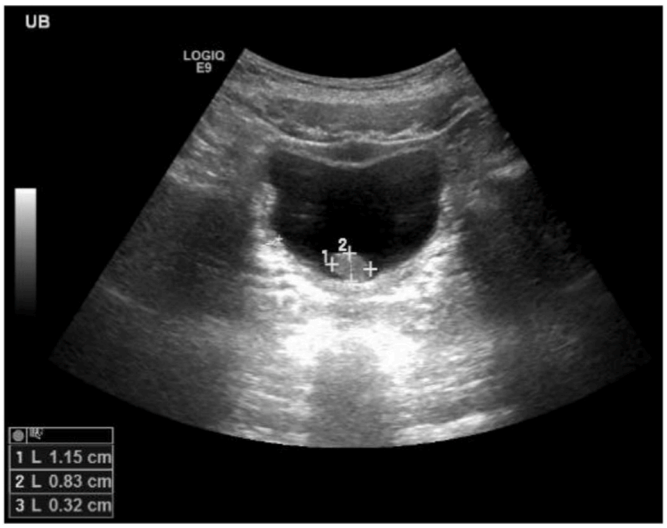

Fig. 1. US urinary bladder showing a 1.2 × 0.8 cm well defined hyperechoic structure in the posterior bladder wall close to the bladder neck.

was a significant post-void residual (PVR) urine volume of 95 ml.

A 17F cystoscope was used for examination, which showed a normal anterior urethra. A protruding polypoidal, circular and narrow neck mass was visible attached to the proximal end of the verumontanum and extended to the bladder neck (Fig. 2). The bladder mucosa showed mild trabeculation without masses or stones. We used a hot knife to reset the mass. A foleys catheter was inserted at the end of procedure and removed on the next day without postoperative complications. The histopathology of the resected tissue confirmed the diagnosis of cystitis cystica and cystitis glandularis (Fig. 3).

One month after the operation, our patient's LUTS improved significantly without further episodes of hematuria. An ultrasound showed no residual mass or significant PVR urine. For further follow-up, our plan is to do ultrasound and cystourethroscopy at 6 and 12 months because the lesion may recur.

Discussion

Urethral cystitis cystica and cystitis glandularis are extremely rare diseases. It is worth noting that there is only one case report describing urethral pseudotumoral cystitis cystica with squamous metaplasia in a 9-year-old boy and there was no histopathological evidence, showing cystitis glandularis.[3] Symptoms of urethral cystitis cystica and cystitis glandulrais are mainly obstructive LUTS (weak stream). Other symptoms may occur, such as frequency, urgency, terminal hematuria, and stress urinary incontinence. Patients usually have normal blood and urine tests. High PVR urine may be the only abnormality.[3] Compared with bladder cystitis cystica, irritative LUTS and hematuria are the main presenting symptoms. Obstructive LUTS is noted in rare circumstances, if the bladder mass is prolapsed causing bladder outlet obstruction.[4] Cystourethroscopy usually reveals a polypoidal mass on the posterior urethra, that may be associated with encrustation and hypervascularity. In long-standing presentation, urinary bladder trabeculations may present. Differential diagnosis includes a urethral fibro-epithelial polyp and rhabdomyosarcoma and it is primarily established by histopathology. The fibroepithelial polyp of the urethra is rare. Hematuria and obstructive LUTS are the most common findings. It consisted of connective tissue and smooth muscle cells covered by a normal transitional epithelium, and sometimes by areas of squamous metaplasia. Rhabdomyosarcoma of the urogenital tract usually originates in the bladder and prostate and is unlikely to appear in the urethra. Histopathology of the embryonal subtype revealed subepithelial aggregates of rhabdomyoblasts "grape-like" tumor cells known as cambium layer.

Transurethral resection of the urethral lesion is considered the only available diagnostic and the therapeutic method.[3] After the urethral mass was removed, the boy reported complete relief of symptoms, and follow-up ultrasound examination showed a decrease in PVR urine. Urethral cystitis cystica and cystitis glandularis may recur, so close follow-up is required. Moreover, the malignant potential of cystitis glandular cannot be ruled out. Filipas and colleagues reported multiple urethral recurrences after resection, and they used systemic corticosteroids to slow down its growth.[3] From an oncology point of view, although cystitis glandularis can coexist with bladder carcinoma as a

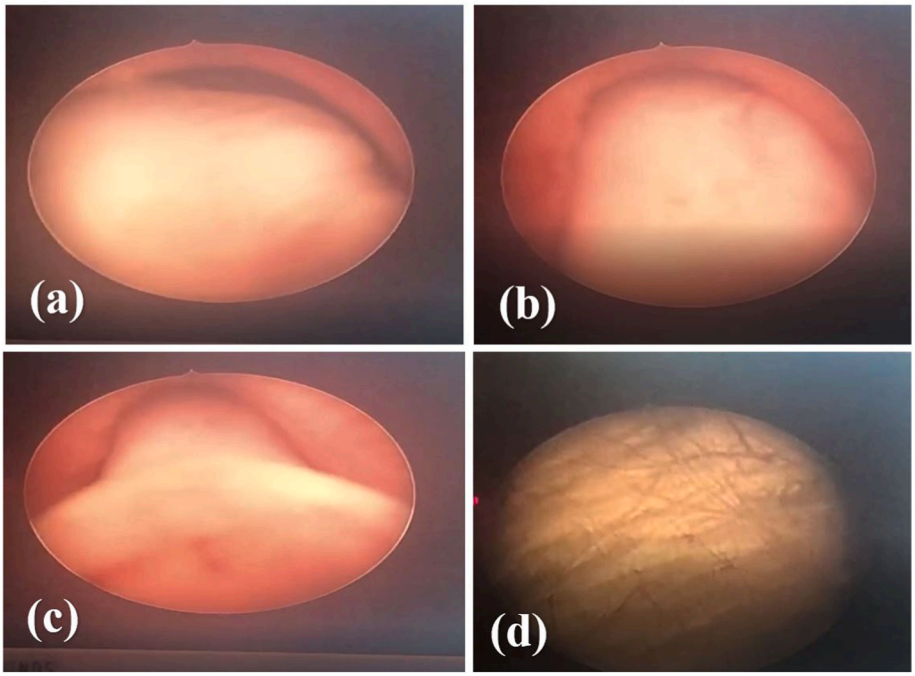

Fig. 2. Cystoscopy showing the obstructing polypoidal mass at the level of bladder neck (a), obstructing the posterior urethra (b), attached to the proximal end of verumontanum (c), and the bladder having mild trabeculations (d).

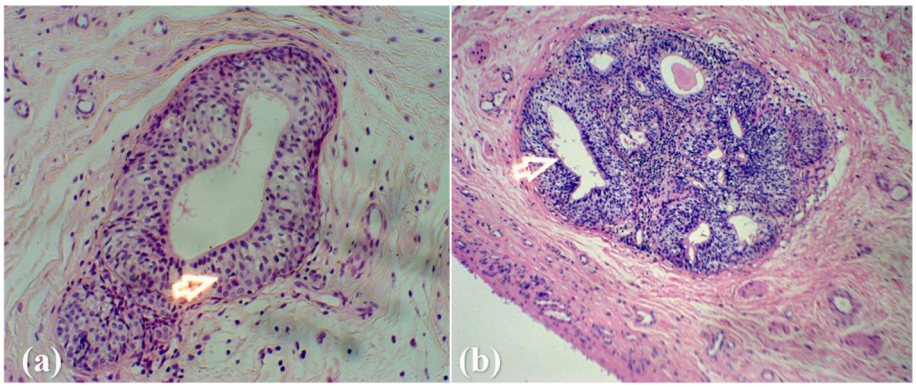

Fig. 3. Showing central lumen lined by glandular columnar epithelium of cystits cystica (a), and a gland in lamina propria lined by columnar epithelium, and the cyst lumen contains esenophilic secretions of cystits glanudlaris of the usual type (b). (Mangification x40, hematoxyline & eosine stain).

pathological finding, there is insufficient evidence in the literature to indicate that cystitis glandularis increases future malignant potential, and its risk before malignant transformation is still controversial.[5]

Conclusion

In summary, urethral cystitis cystica and cystitis glandularis are rare diseases, presented with obstructive LUTS and terminal hematuria. Patients may have high PVR urine volume. Transurethral visualization and resection of lesions is diagnostic and therapeutic. Close follow-up is mandatory because recurrence may occur.

Consent

Written informed consent was obtained from the patient.

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Declaration of competing interest

All authors have no competing interest related to this study.

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