Case Report

Ureteritis cystica: A rare entity in children

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ABSTRACT

Ureteritis cystica (UC) is a benign condition. Although it can often be diagnosed with imaging techniques, we report a case of a child for whom we planned nephrectomy and ureteral augmentation cystoplasty, but abandoned the cystoplasty due to extensive UC in the ureter.

Key words: Augmentation cystoplasty, pathology, ureteritis cystica

INTRODUCTION

Ureteritis cystica (UC) is a benign pathology, which develops following chronic inflammation. A variety of diseases with obstruction and chronic inflammation may lead to UC. Although a diagnosis is often made with imaging techniques, we report a case of a child in whom was discovered during a nephrectomy. To the best of our knowledge, this is the first pediatric case of UC.

CASE REPORT

The patient was a 12 year old child who, as a newborn, had undergone surgery for anal atresia with rectourethral fistula repair and colostomy. His postnatal ultrasound (US) and Dimercaptosuccinic acid (DMSA) scan had shown a hypoplasic left kidney and bilateral minimal hydronephrosis. Voiding cystourethrography (VCU) showed bilateral grade 5 vesicoureteral reflux. At 1 year of age, the colostomy was closed and DMSA was repeated and differential function of the left kidney was found 22%, whereas the right kidney was 78%. In the 3rd year of life, a second VCU revealed clinically significant residual urine, which required clean intermittent catheterization (CIC). He underwent bilateral Cohen ureteroneocystostomy for recurrent urinary tract infections (UTIs) under antibiotic prophylaxis. Ultrasound prior to surgery showed bilateral high grade hydronephrosis. On the follow-up, DMSA scan showed 63% differential function on the right kidney. At the age of 7, US showed persistent bilateral high grade hydronephrosis. When he became a 10-year-old, hemodialysis was initiated for the first time and he had been placed on a renal transplant waiting list. The patient was on CIC 6 times a day when another urodynamic evaluation was obtained at 12 years of age. The study revealed hypocompliant bladder with decreased anatomic capacity accompanying detrusor overactivity with no demonstrable vesicoureteric reflux. Due to the low functional bladder capacity, nephrectomy with ureteral augmentation cystoplasty was planned.

During surgery, after left simple nephrectomy, left ureter was dissected and spatulated, but ureteral mucosa was found to be filled with polyps [Figure 1]. It was decided not to use the ureter and the augmentation cystoplasty was postponed.

Histopathological examination of the ureter showed polypoid ureteritis and exuberant UC producing exophytic mucosal lesions in a background of chronically inflamed lamina propria [Figure 2]. UC typically consisted of expanded cystic lesions containing colloid like eosinophilic fluid. Cysts were lined by flattened urothelial cells without atypia.

DISCUSSION

Ureteritis cystica is a rare and benign pathology of inflammatory origin first reported by Morgagni in 1761 and described by

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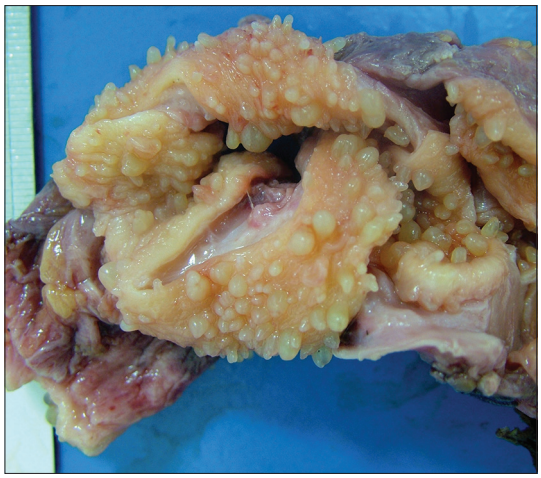

Figure 1: Innumerable 2-4 mm pearly raised vesicular/polipoid lesions covering entire mucosa of the ureter

Richmond and Robb in 1967.[1] It is the cystic transformation of von Brunn's nests.[2] A PubMed search with keywords of "ureteritis, cystica" revealed 72 papers those mostly presenting case reports without any pediatric age patients. Although many reports state female dominancy, Menendez *et al.* have reported this may not be true after evaluating 34 cases and showing only 44% of them being female patients.[3]

Ureteritis cystica is associated with chronic irritation. Some studies have shown nephrolithiasis and recurrent UTIs to be the cause. It is most frequently diagnosed incidentally.[1] UC is generally seen in patients during the evaluation of UTI (82%), urolithiasis (53%), and hematuria (52%).[4] Furthermore, it can present as renal colic. The differential diagnosis includes ureteral tumors, pseudodiverticula, urolithiasis, polyps, vascular indentations, tuberculosis, UTIs, and submucosal hemorrhages.[1] In the long-term, UC may result in an atrophic kidney due to obstruction.

In this particular case, the etiology was thought to be persisting recurrent UTIs initially due to the bilateral high grade reflux and then due to the obstruction that developed following antireflux surgery. Mahboubi *et al.* have reported UC in a patient who received formalin treatment for cyclophosphamide induced hemorrhagic cystitis.[5] Although, intravenous pyelography and retrograde urography were the preferred diagnostic tools earlier, in the last decade magnetic resonance and computed tomography urography have become more available and accurate. Despite advanced technology, imaging studies may be inadequate in some cases. Ureteroscopy is a definitive diagnostic approach in these cases[2,3] In the present case, we did not have any imaging studies other than US since such pathology was not expected. Moreover, the patient's chronic renal failure precluded the use of imaging studies with contrast agents. Therefore, in such patients with history of chronic UTI, inflammation or obstruction, the possibility of this pathology should be taken into consideration and appropriate imaging

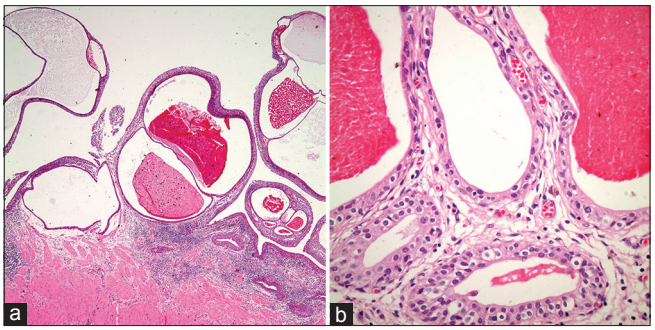

Figure 2: Florid ureteritis cystica with chronic inflammation in lamina propria. Large and small cysts lined by flattened urothelial cells leading to polipoid mucosal protrusions into the lumen; some filled with eosinophilic secretion ((a) H and E, ×40; (b) H and E, ×400)

modalities such as retrograde ureteropyelography or direct examination by ureterorenoscopy may be needed before reconstructive surgeries using the ureter. Limited data are present in the literature regarding follow-up and malignant transformation, however follow-up is advised annually. Duffin *et al.* have reported a case after follow-up of 17 years without malignant transformation, whereas there is a report of a case complicated with adenocarcinoma of the ureter.[4]

During the surgery we abstained from using this ureter as we did not have the facility of frozen section pathology and also had no experience of using such a ureter for augmentation. No bladder biopsies were taken during or after the surgery. The treatment of this pathology, which is believed to be reversible, depends on the elimination of the underlying causes.

CONCLUSION

Ureteritis cystica is a rare pathology that should be considered in the presence of chronic inflammation and obstruction of the upper urinary tract. Presence of such pathology needs to be considered in cases undergoing ureterocystoplasty.

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