A Case of Calcified Ureteritis Cystica: An Indiscernible Condition from Ureterolithiasis

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Abstract: Ureteritis cystica is characterized by formation of multiple cysts in the wall of the renal pelvis or ureter. The clinical course is usually slow, but manifests if the cysts cause infection or obstruction. Stones are further complication to the disease. In this case study, we present a 39-year-old male originally referred with renal colic and misdiagnosed as ureterolithiasis due to the calcifiying cysts.

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Introduction

Ureteritis cystica or cystic pyeloureteritis is a rare benign disease that has been known since 1761; however, the etiopathogenesis is still unclear. This condition is characterized by the formation of small cysts on the submucosa of the collecting system. The disease is seen mostly in the middle to advanced aged patients and the course is indolent but almost always associated with persistent or recurrent urinary infections. The radiological findings typically include numerous small polypoid filling defects in the collecting system, mostly without obstruction [1–8]. Calcification of the cysts has not been reported previously in the literature. Herein, we present a case of ureteritis cystica consisting of calcified cysts.

Case

A 39-year-old male was admitted to a secondary health care institution complaining of right flank pain and dysuria lasting for ten days. Urine analysis revealed hematuria and proteinuria and no radioopaque calculi were seen on direct roentgenograms. Intravenous urography (IVU) revealed a delay in the nephrogram and pyelogram phases and hydronephrosis of the right kidney, suggesting obstruction. However, since delayed urographies were not taken, the right ureter was not visualized (Figure 1). The patient was diagnosed as having nonopaque ureteral calculus and was prescribed antibiotics and spasmolytic drugs. Since the symptoms did not improve in spite of medical treatment, the patient was referred to our Urology department. Computer tomography without contrast injection was performed and showed the site and cause of the obstruction, which were bilateral small calculi in both kidneys. Right hydroureteronephrosis was also detected. Three distinct radiodense lesions were located at 1 cm intervals at the distal part of the right ureter with periureteral inflammation (Figure 2). Magnetic resonance urography (MRU) revealed a signalvoid lesion causing obstruction (Figure 3). A diagnosis of Ureterolithiasis was done, and surgery was planned. Unexpectedly, a cystic lesion was seen on exploration (Figure 4) and the proximally located cyst was excised.

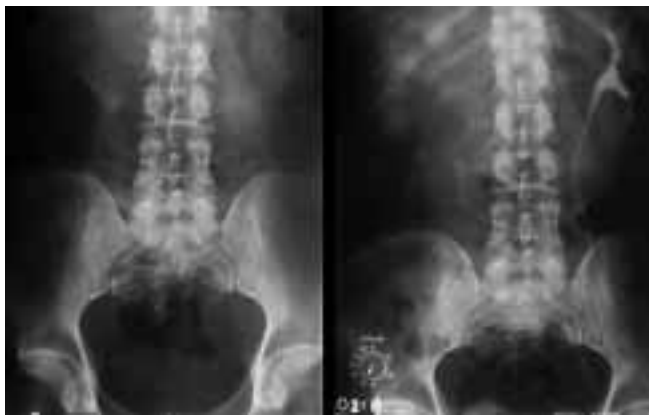

Figure 1 – Urographies done in another health institute. No radioopaque lesion was detected on direct roentgenogram (right). Intravenous pyelography (left) revealed delay of pyelogram phase and hydronephrosis of right kidney indicating some obstruction. But since delayed graphies were not been taken ureter was not been visualized.

Histopathologically, active chronic inflammation was reported. Furthermore, as the patient improved, it was not necessary to perform a follow-up IVU.

Discussion

Ureteritis cystica is a benign chronic disease, which is usually found with urinary tract infections, urolithiasis, and hematuria due to stasis of urine. As a result of the inflammatory reaction, localized fibrosis forms around the cysts [1–8]. Obstruction occurs as a result of projectile cysts or localised fibrosis surrounding the Brunn's

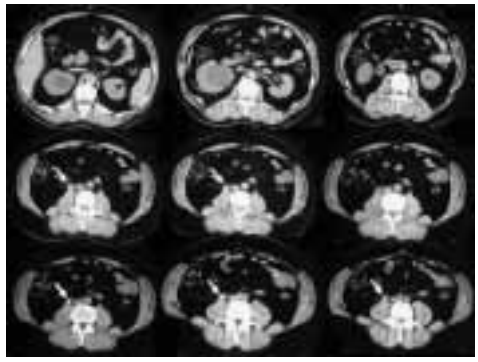

Figure 2 – Computed tomography without contrast injection. Right hydroureteronephrosis as well as small calculi in the left kidney (B). Radioopaque lesions (white arrows) in ureteral lumen associated with periureteral inflammation are seen.

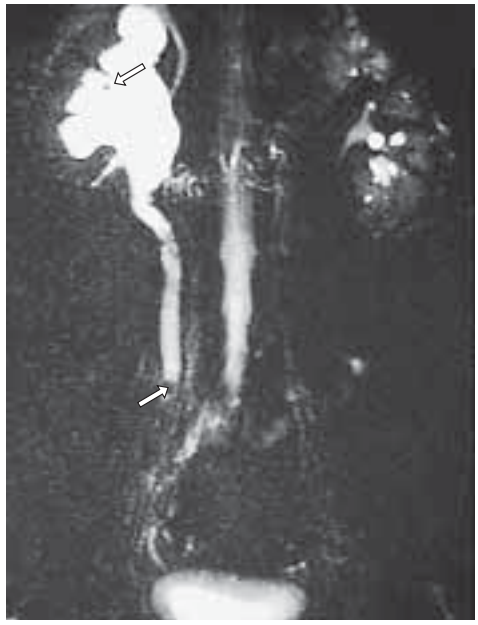

Figure 3 – Magnetic resonance urography depicts signal-void lesion in distal 1/3 ureter causing obstruction (the other two lesions are not seen). A small calculus is seen in right kidney (small arrow).

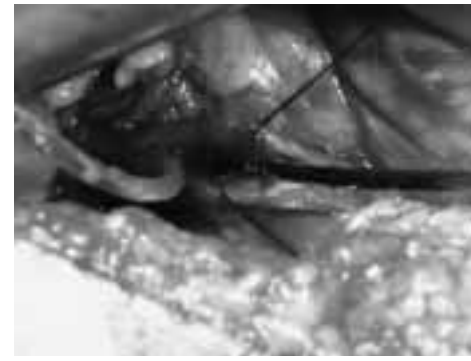

Figure 4 – Calcified cyst was unexpectedly found during the exploration.

nests and small cysts [3]. Notably, chronic irritation can continue for years [2] and only one case of malignancy (adenocarcinoma) with ureteritis cystica has been reported to date [9].

Although the reported cases of ureteritis cystica suggest female and older patient dominance [2, 5, 6], a review by Menendez [4] consisting of 34 patients reveals a slight male (56%) majority and a range of 30–77 (mean 59) years of age. The cysts can be located within the excretory system but mostly they occur at the ureter (79%), chiefly over the upper third [1]. Bilateralism was reported in 20.5% of the patients in the review [4].

Radiological diagnosis includes multiple small filling defects throughout the collecting system. Unfortunately, the urographies of our case were incomplete and traditional retrograde pyelography better demonstrates filling defects, especially if obstruction exists [1–8]. However, sectional imaging modalities are also capable of showing the level and cause of the obstruction non invasively. The radiologic findings were not typical for ureteritis cystica in our case. On IVU, the ureter was not adequately filled with contrast material, subsequently the filling defects were not obvious. Retrograde pyelography could better visualize the filling defects but since the stone was suspected, we preferred non invasive, cross sectional imaging techniques.

The differential diagnosis of filling defects in pyelography includes: clots, air bubbles (for retrograde pyelography), stones, ureteral polyps, transitional cell carcinoma, malacoplakia, virus induced ureteric strictures, sloughed renal papillae in diabetes or analgesic nephropathy or mucosal changes in schistosomiasis, tuberculoma, fungus [4], epidermoid cysts [10–13], and extrinsic indentations due to vascular compression [4].

Other cystic lesions of the ureter are epidermoid cysts which are extremely rare in the urinary tract [10–13]. These cysts may mimic ureteral stones clinically (renal colic, flank pain, microscopic or macroscopic haematuria) and can be seen radiologically. Ishizaki and Gokce [10, 12] reported calcified epidermoid cyst cases, which are impossible to distinguish from ureteral stones radiologically.
The presence of keratinized material in the urine sample supports the epidermoid cysts. In our case, the diagnosis of epidermoid cyst was also excluded by the immunohistochemical study.

Tuberculoma is the other cause of calcified lesions in the ureter, and absence of the other findings of tuberculosis excluded this diagnosis in our case. Ureteral polyps and transitional carcinoma could be diagnosed pathologically [4, 10].

A specific treatment for ureteritis cystica is still lacking. Dilation of the ureter, mechanical rupture of the cysts and instillation of silver nitrate has all been tried but are no longer used. The associated pathologies (obstruction, calculus, infection) should be treated in symptomatic cases. However, there is no agreed therapy protocol for asymptomatic cases [4]. Endoscopic resection of the cysts is thought to be the treatment of choice, but unfortunately since we do not have an ureteroscope, we had to perform open surgery.

To the best of our knowledge, ureteritis cystica with calcified cysts has never been reported before. The clinical and radiological features may mimic ureteral stones, moreover, they can be found with stones-making a preoperative diagnosis impossible.

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