Renal papillary hypertrophy, a rare cause of recurrent gross hematuria; Case report and review of literature

Ahmad Beltagy, Mohamed Elsaqa, Islam Koraiem, Ahmed Abulfotooh Eid

Department of Urology, Faculty of Medicine, Alexandria University, Alexandria, Egypt.

Summary
Hematuria is a critical symptom that should properly be investigated. One of the rare causes is renal papillary hypertrophy. Literature review revealed only few reported cases. Biopsy in reported cases has shown hyperplasia of renal papillae with normal histology. We report a case of bilateral renal papillary hypertrophy in a 32 years old female presented with intermittent gross hematuria. Computed tomography urography, cystoscopy and selective cytology did not show any positive findings. Retrograde flexible ureterorenoscopy showed enlarged renal papillae protruding into upper and middle calyces of both kidneys with clots and active bleeding in some. Holmium:YAG Laser ablation of hypertrophic papillae showed an effective minimally invasive management of the condition.

Key words: Renal papillary hypertrophy; Hematuria; Laser ablation; Flexible ureteroscopy.

Submitted 1 May 2020; Accepted 6 June 2020

Background
Hematuria, either gross or microscopic, should be carefully investigated. Hematuria, specifically in adults, should be regarded as a symptom of urologic malignancy till proved otherwise. All patients with hematuria, except perhaps young females with acute bacterial hemorrhagic cystitis, should be thoroughly evaluated (1). Renal papillary hypertrophy or hyperplasia is one of the rare causes of gross or microscopic hematuria of benign origin. Few cases were reported in the literature (2–7). In this article, we report this case presented with gross hematuria due to bilateral renal papillary hypertrophy and its minimally invasive management with Holmium:YAG laser ablation together with literature review of this rare pathology and its management.

Case report
A 32 year old female presented with intermittent total gross hematuria with occasional clots and bilateral loin pain for 6 months. Other than history of poliomyelitis affecting her left leg with scoliosis of the spine, the patient has no evident clinical history or abnormal clinical findings. She did not complaint of bleeding elsewhere. Also, she did not receive neither anticoagulant nor anti-platelet medications. Clinical examination showed normal blood pressure. Urine analysis revealed hematuria (> 100 red blood cells) without pyuria or proteinuria. Blood count, renal and liver functions and bleeding profile were normal. Urine culture and ultrasound were unremarkable. Multiphasic renal computed tomography (CT) showed bilateral enlarged papillae causing concave impressions in the calyces of both kidneys, a picture suspicious of hypertrophied renal papillae (Figure 1). Diagnostic cystoscopy with barbotage bladder cytology were done; both failed to show any positive finding. Diagnostic retrograde intrarenal ureteroscopy was done using 7.5 Fr flexible ureteroscopy that revealed hypertrophied renal papillae in the upper and middle calyces of both kidneys with active bleeding in some of these papillae (Figure 2). Selective cytology of both ureters revealed no suspicious findings.

In a later setting, Holmium:YAG laser ablation of hypertrophied papillae of both kidneys was done using 7.5 Fr flexible ureteroscopy and 365 μm fiber at settings of 1200 mJ and 12 Hz. After 3 months follow up, the patient did not report any attacks of gross hematuria, despite the persistence of microscopic hematuria. She also described significant improvement of bilateral loin pain. Patient was consented for further process of data collection and publication.

Discussion
Renal papillary hypertrophy is one of the rare causes of hematuria. Only few cases have been reported in the literature. In 1956, Laurent first reported renal papillary hypertrophy in a 9-month-old female infant after nephrectomy for suspected pelvic tumor by retrograde filling defect (2). In 1961, Moonen et al. described two cases of a 25-year-old female with microscopic hematuria and 11-year-old male with loin pain (3). Histopathology following nephrectomy in these two reports revealed hyperplasia of renal papillae with normal histology (2, 3).

The same pathology was reported later on by only few case reports (4-7). The age of reported cases ranged between 9 months and 30 years old. To our best of knowledge, only 8 cases were reported in the literature, 6 females and 2 males. Presentations are usually gross or microscopic hematuria with or without flank pain (2-7). However, although association with use of oral contraceptive pills was seen in 3 reported cases, our case never

No conflict of interest declared.
used oral contraceptive pills (6, 7). Histopathology, either after nephrectomy or recently with endoscopic biopsy, showed enlarged hyperplastic papillae (multiple rows of cells) with normal histology associated with dilated thin walled veins at the angles of calyces which may be responsible for bleeding (2, 3). CT Urography, retrograde study or intravenous pyelography may show lobulated filling defects. Magnetic resonance (MR) Urogram in 2 reports showed prominent medullary pyramids with apices protruding into calyces causing enlargement and deformity at the calyceal fornice (5, 6). Diagnosis was previously reported after nephrectomy for suspected renal or urothelial tumors caused by filling defect on intravenous pyelography or retrograde study. Recently, diagnosis depend on direct visualization of hypertrophied papillae by retrograde flexible ureterorenoscopy. Biopsy may be omitted, as in our case, in case of multiple or bilateral lesions, negative cytology and absence of suspicious findings. There are no definite guideline recommendations regarding management of papillary hypertrophy. Birk et al. reported the effective use of Holmium:YAG laser for ablation of hypertrophied papillae in 2 cases (6). Heißler et al. suggested the use of either Holmium-YAG or Thulium-YAG laser for ablation according to their use for ablation of upper tract urothelial tumors (7). In our case, we used 365 µm Ho:YAG laser fiber for bilateral ablation.

**Conclusions**
Renal papillary hypertrophy is a rare cause of gross or microscopic hematuria of that should be kept in mind as differential diagnosis for recurrent hematuria of benign origin. Retrograde intrarenal laser ablation represents the most appropriate management.

**References**