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DIFFUSE PAPILLOMATOSIS OF THE UPPER URINARY TRACT: A RARE MIMICKER OF UPPER TRACT UROTHELIAL CARCINOMA: A CASE REPORT

Urology

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ABSTRACT

Diffuse papillomatosis is a rare, malignant growth in the urinary tract with unknown etiology, with only two cases reported to date globally. We report the first case of diffuse papillomatosis affecting the urinary tract from Asia in a 58-year-old healthy woman who presented with a one-week history of painless hematuria. CT scan revealed heterogeneously enhancing lesion in the right kidney with fleshy central calyceal growth of 1.5cm diagnosed by ureteroscopy, but with no evidence of any invasive malignancy. The patient underwent a right robotic nephroureterectomy. Clinically and radiologically, it mimics upper tract urothelial carcinoma that can be mistakenly diagnosed as cancer and is reported for its rarity.

KEYWORDS

Diffuse papillomatosis, urothelial papilloma, robotic nephroureterectomy, upper tract urothelial carcinoma

INTRODUCTION

The urothelial papilloma is usually a smooth polypoid lesion that possesses a stalk. Diffuse papillomatosis indicates the presence of numerous papillomas and its tendency to disseminate urinary passages[1]. Diffuse papillomatosis is a rare entity about which only two cases have been reported in the literature.

Case History

A 58-year-old, healthy mother of two presented with a history of gross, total, painless haematuria for one week. She had undergone two lower section caesarean sections (LSCS) and a vaginal hysterectomy for uterine fibroids ten months earlier. She had no contributing family history, history of smoking or exposure to any carcinogen. Her initial clinical examination was unremarkable, and pelvic ultrasound revealed moderate right hydronephrosis.

Preliminary blood results were within normal limits, but with mild iron deficiency anaemia, and urine routine, showed 10-15 RBC/HPF. A contrast-enhanced computed tomography (CT) scan revealed a heterogeneously enhancing lesion with areas of haemorrhage in the mid pole calyceal system of the right kidney with extension into the renal pelvis [Fig. 1]. Nodular wall thickening and enhancement of the right renal pelvis, proximal ureter and right vesicoureteral junction (VUJ) were also noted. Urine cytology revealed atypical urothelial cells (AUC), according to the Paris system for reporting urine cytology. A diagnostic ureteroscopy and cystoscopy revealed a fleshy middle calyceal growth of 1.5 cm with slough noted projecting into the pelvis along with erythema of the pelvis surrounding the lesion with a grossly normal ureter.

The patient underwent a right robotic nephroureterectomy. Pathology revealed diffuse papillomatosis of the ureter and the renal pelvis with no evidence of any invasive malignancy (Fig. 2). In the third month follow up, the patient is doing well with no further haematuria and no evidence of recurrence on cystoscopy.

DISCUSSION

Urothelial papilloma usually occurs in the bladder, with the highest incidence in the trigone and bladder neck locations2. The presence of papilloma is noted in the other parts of the urinary tract, including the pelvis, ureters and urethra3. Diffuse papillomatosis is a rare disorder in which papillomas occur extensively in the urinary tract. The present case of diffuse papillomatosis extending from the renal pelvis running down to the entire length of the ureter is quite unusual. It is the third report of diffuse papillomatosis of the upper urinary tract in literature and the first case of isolated upper tract involvement. Rayer4 in 1841 and Cecilvillaunte1 in 1952 reported diffuse urothelial papilloma in women of age 58 and 67, respectively. The presentations of the cases were similar to ours.

The aetiology of the papillary growth in the epithelium in the pelvis disseminating into the entire length of the ureters is not clear. Cecilvillaunte1 discussed the possibilities of certain substances linked with bladder cancer like aniline-dye, beta-naphthylamine and other carcinogenic irritants of the epithelium. Nevertheless, the present patient was not exposed to any of these risk factors.

In the present case, the radiological imaging, endoscopic visualisation and urine cytology were more suggestive of upper tract urothelial carcinoma (UTUC). Since UTUC is highly malignant, an aggressive radical nephroureterectomy was performed. Histology showed a typical benign fibrovascular core with multiple overlying layers (up to 7 layers) with a normal urothelial lining, extending from renal pelvis throughout the ureter(Fig. 2). There was no cellular atypia or loss of polarity. Diffuse papillomatosis is a benign entity that resembles multifocal urothelial carcinoma, and usually, it is considered a histological surprise.

CONCLUSIONS

Limited research exists to comment on the management and follow up of the diffuse papillomatosis patients. The present case is an exclusionary diagnosis and should be considered as a mimicker of UTUC. The case is reported for its rarity.

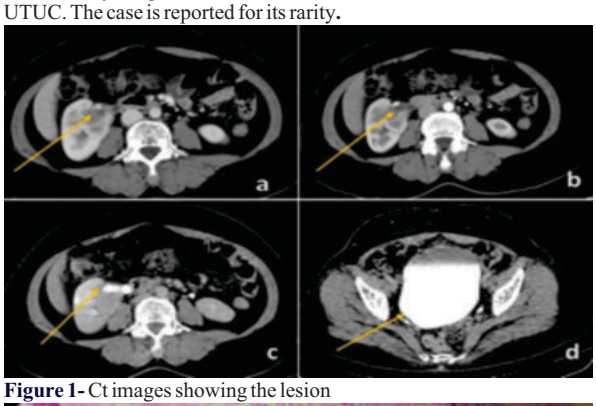

Figure 1- Ct images showing the lesion

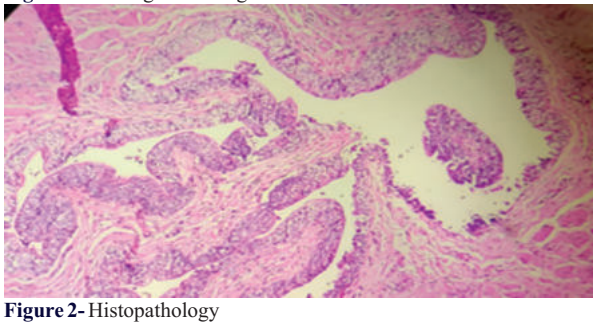

Figure 2- Histopathology

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