Adenomatous Metaplasia Arising in a Horse-Shoe Kidney - A Rare Entity

Sukhpreet Kaur¹ · Shariq Ul Hasan²

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Abstract Urinary adenomatous metaplasia (nephrogenic adenomas) is uncommon, benign, apparently metaplastic response of the urothelium to chronic inflammation, trauma, surgery, urolithiasis, long-term indwelling urethral catheterization, and immunosuppressive therapy instillation. Gross or microscopic hematuria and some irritable urinary tract symptoms are often observed. A diagnosis is primarily based on the histopathological examination. We are reporting this case because adenomatous metaplasia is very rarely reported in the kidney especially in a horse shoe kidney with a large renal stone and these lesions could be mistakenly diagnosed as malignancy. In our knowledge this is the very first case of its kind. An awareness of this entity is critical to prevent over-diagnosis of cancer and to avoid unnecessary treatment.

Keywords Horse-shoe kidney · Metaplasia · Nephrogenic adenoma · Urothelium

Introduction

Adenomatous metaplasia (AM) is a rare metaplastic benign lesion of urothelium in response to genitourinary tract procedures, chronic infection, trauma, urinary calculi and immunosuppressive therapy after renal transplantation [1]. It has been reported to occur throughout the urinary tract from the renal

pelvis through the urethra. The most frequent location is the urinary bladder. It rarely if ever arises in the absence of a preexisting urogenital abnormality. Adenomatous metaplasia may resemble certain types of urologic malignancies [2]. In our knowledge this is the very first case of its kind.

Case History

This is a detailed clinical and histopathologic report of an adenomatous metaplasia arising in the renal pelvis of a 48-year-old man. The patient presented with a complaint of right flank pain, two episodes of painless hematuria & dysuria. Routine lab investigations were within normal limits. Contrast enhanced CT scan abdomen & pelvis was done. It revealed horseshoe shaped kidney with a 2.5 cm calculus in right renal pelvis causing gross dilatation of pelvicalyces. Thinned out renal parenchyma with no excretion of contrast was noted. Left kidney show normal excretion of contrast. Isotope renal scan also shows right non functioning kidney. A diagnosis of right nonfunctioning kidney was made and simple nephrectomy was performed.

Grossly, the surface of kidney was irregular with adherent capsule. On cutting; markedly dilated pelvi-calyceal system was noted (Fig. 1). Cortex was atrophied and the corticomedullary junction was difficult to discern. Very fine minute papillary excrescences were seen in the pelvis. No obvious growth was identified. On light microscopic examination, sections revealed a metaplastic epithelium lining the pelvi-calyceal system. The epithelium was thrown into papillae and tubules. These were lined by single layer of bland cuboidal cells or hobnail cells (Fig. 2).

These cells had clear or eosinophilic cytoplasm, small nuclei and inconspicuous nucleoli. Some of the cells and tubules showed eosinophilic secretions. Mild to moderate anisonucleosis was seen. Minimal atypia and few mitotic figures were noted. There was no evidence of desmoplasia. In

✉ Sukhpreet Kaur
drsukh_kaur@yahoo.co.in

¹ Department of Pathology, Bhopal Memorial Hospital and Research Centre, Under Indian Council of Medical Research (ICMR), Raisen Bypass Road, Bhopal, M.P., India

² Department of Urology, Bhopal Memorial Hospital and Research Centre, Under Indian Council of Medical Research (ICMR), Raisen Bypass Road, Bhopal, M.P., India

Fig. 1 Cut surface of kidney showing very fine and minute papillary excrescences in the pelvis. In the inset, dilated pelvicalyceal system, atrophied cortex and no demarcation of cortico-medullary junction is seen

some areas, pelvis was lined by benign normal to attenuated transitional epithelium. No invasion was seen. On Immunohistochemistry, the cells were positive for Pancytokeratin and negative for vimentin. Ki-67 index was <1 %. Rest of the areas reveal global glomerulosclerosis and obsolescence. Tubular damage and thyroidisation was evident. Stroma reveal foci of xanthogranulomatous inflammation and focal lymphoid aggregates. A diagnosis of adenomatous metaplasia with xanthogranulomatous reaction and chronic pyelonephritis was made. Two years postoperatively, the patient is doing well.

Discussion

Urinary adenomatous metaplasia, also known as nephrogenic adenoma, is a rare condition of the urinary tract. The lesion was first described by Davis in 1949 and Friedman and Kuhlénbeck coined the term nephrogenic adenoma in 1950.

Fig. 2 Section from pelvis showing adenomatous metaplasia. The epithelium is thrown into papillae and tubules. These are lined by cuboidal epithelium (H&E, 20X). Pan-CK positive cells in the inset

[2, 3] The terms nephrogenic metaplasia [4] and adenomatous metaplasia [5] are preferred by some. It has been reported throughout the entire urinary tract. The most frequent location is the urinary bladder. However, in our case the lesion was in the renal pelvis and the rarity of the lesion in the kidney makes it unique.

There are two different opinions on its histogenesis. The prevalent one was that nephrogenic adenoma was a metaplastic change of *de novo* urothelium in response to persistent irritation, recurrent infection or trauma, calculi, anatomic defects, trauma, surgery, benign or malignant tumors of the lower urinary tract, and intravesicular immunotherapy such as BCG or chemotherapeutic agents. Ford et al. in their study with 70 cases pointed out that lesions occurred mostly at the site of previous operations. [5] The other theory postulated on embryologic origin involving remnants of the mesonephric duct as is illustrated by Bhagavan et al. [6] and Mazal et al. [7] In our case, the kidney was horse-shoe shaped and had a large renal stone. We think that both metaplastic theory as well as embryologic derivation from mesonephric remnants is responsible for such lesions. As there is adenomatous metaplasia of the transitional epithelium in our case, the descriptive term “adenomatous metaplasia” seems more appropriate than the designation “nephrogenic adenoma” especially in case of renal lesions.

On light microscopy, AM shows a variety of patterns [8, 9] including tubulocystic, papillary, and much less frequently solid, that often coexist. Recognition of its characteristic patterns may help in making the diagnosis and distinguishing this lesion from malignant neoplasms occurring at the same sites. Most tubules, cysts, and papillae have cuboidal to low columnar epithelium with scant to abundant eosinophilic cytoplasm. These tubules are similar to mesonephric tubules, usually lined by single layer of bland cuboidal or hobnail cells, surrounding eosinophilic or basophilic secretions [9]. Sometimes tubules are surrounded by a prominent basement membrane. It may involve lamina propria but spares muscularis propria. Occasionally luminal blue mucin compresses the nuclei giving a signet ring-like appearance. The nuclei are regular and round with no prominent nucleoli and rare atypia [9]. Mitotic figures are absent or rare. Stroma contained inflammatory infiltrate. No desmoplasia was seen. Although AM are considered to be benign lesions, malignant transformations have been reported suggesting it a premalignant disease. Approximately 90 % of patients with adenomatous metaplasia are adults, and there is a male preponderance of 2:1. In children, it is more common in girls than in boys. Complaints of hematuria, dysuria, and frequency are common.

The nephrogenic adenoma with predominant papillary pattern may cause confusion with papillary urothelial carcinoma. Recognition of the cuboidal epithelium covering the papillae of nephrogenic adenoma differentiates it from other lesions,

which are covered by urothelium. It may cause confusion with adenocarcinoma especially when it involves urinary bladder or prostate. In contrast to adenocarcinoma, adenomatous metaplasia lack solid growth patterns, mitotic activity, prominent cytological atypia, and are not typically large deeply invasive tumors. Clear cell adenocarcinoma of urinary bladder can be confused with adenomatous metaplasia. To conclude, we must correlate microscopic as well as gross findings with clinical history for the diagnosis of AM. An awareness of this entity is critical to prevent over-diagnosis of cancer and to avoid unnecessary treatment.

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