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Intestinal-type adenocarcinoma arising from a villous adenoma in the renal pelvis in the context of a kidney abscess: Case report

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ABSTRACT

Villous adenoma is a benign neoplasm with an exceptional presentation in the renal pelvis, hence very few cases have been reported. Herein we present the case of a patient who presented with left flank pain clinically suggestive of complicated pyelonephritis, culminating in simple nephrectomy with a villous adenoma in the renal pelvis as histopathological finding associated to the presence of a microscopic focus of intestinal-type adenocarcinoma.

1. Introduction

Villous adenoma is an infrequent benign neoplasm of the urinary tract, the urinary bladder being the most common site of presentation; however, finding it in the renal pelvis is exceptional, with only a few cases reported in the English literature. It is histologically indistinguishable from its colorectal counterpart.[1–3] In addition, up to 35% of these cases may have an associated invasive adenocarcinoma, so an intentional search by the surgical pathologist to rule it out is mandatory in these cases.[2]

2. Case

A 72-year-old female presents with left flank pain of two months duration, attenuated with position changes, accompanied by fever and polyuria. She is hospitalized and antibiotic treatment is started under the clinical suspicion of complicated pyelonephritis. During the approach, a computed tomography (CT) scan with intravenous contrast was performed. It revealed a left renal abscess, hydronephrosis, and a staghorn calculus, for which percutaneous drainage was placed (Fig. 1A). As there was no clinical improvement, coupled with the formation of a cutaneous fistula draining purulent fluid, a simple nephrectomy was performed, but was complicated due to the presence of multiple peritoneal adhesions at the level of the splenic flexure of the colon, so during the procedure it was decided to perform splenectomy and partial colectomy as well. Gross examination revealed a terminal kidney, a staghorn calculus occupying the renal pelvis, purulent fluid, and multiple exophytic lesions with a papillary appearance, which extended close to the ureteral surgical border (Fig. 1 B).

Histopathological findings were villoglandular fronds lined by pseudostratified columnar epithelium, in addition to intestinal metaplasia in the renal pelvis and ureter with involvement of the ureteral surgical border. Another finding was of intestinal-type adenocarcinoma with microinvasion to the lamina propria (Fig. 2). Immunohistochemistry (IHC) stains were performed for CK20, CDX2, and SATB2, all of which were positive and corroborated the intestinal differentiation of the tumor (Fig. 3).

After five days of favorable clinical evolution during the patient's postoperative period, hospital discharge was decided. After one year of follow-up, the patient is asymptomatic, in good general condition, and with no evidence of distant metastasis.

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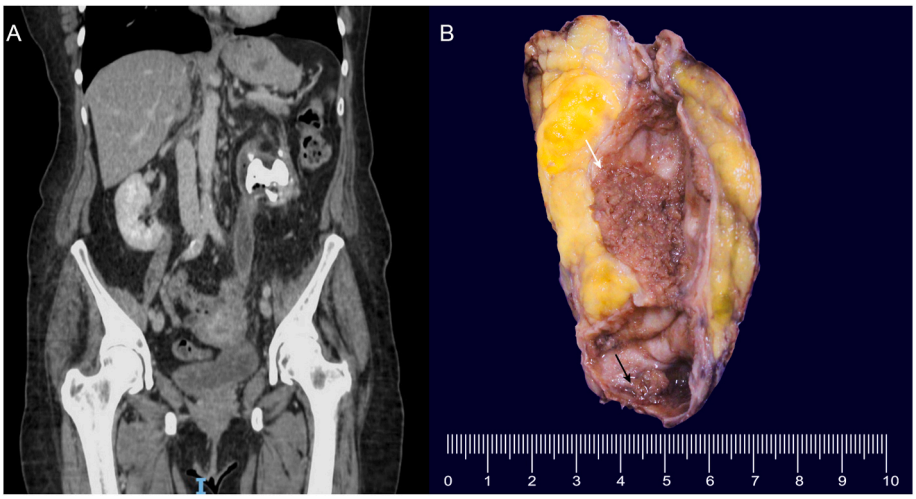

Fig. 1. A. Contrast-enhanced CT showing findings consistent with a left renal abscess, hydronephrosis, and ipsilateral staghorn calculus. B. Product of simple left nephrectomy with loss of normal renal architecture, dilation of the pyelocaliceal system, and presence of exophytic lesions with papillary appearance in the renal pelvis (white arrow) that macroscopically extend close to the ureteral surgical border (black arrow).

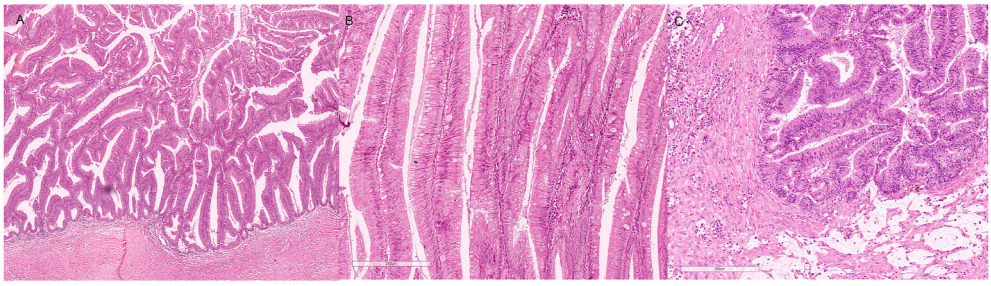

Fig. 2. A. Villoglandular fronds lined by pseudostratified columnar epithelium (H&E, x10). B. Foci of intestinal metaplasia present in the urothelium of the renal pelvis and ureteral surgical border (H&E, x10). C. Microscopic focus of intestinal-type adenocarcinoma with infiltration of the lamina propria (H&E, x10).

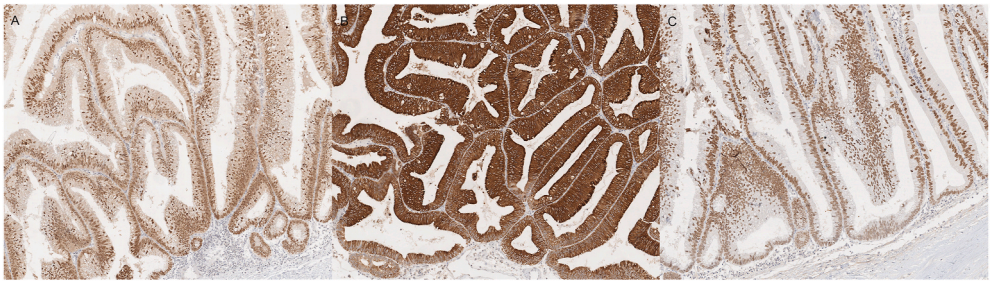

Fig. 3. Immunohistochemistry (IHC). A. Nuclear positivity for CDX2. B. Cytoplasmic positivity for CK20. C. Nuclear positivity for SATB2.

3. Discussion

Described frequently as a benign neoplasm, villous adenoma most commonly arises in the gastrointestinal tract, but it can rarely be found throughout the urinary tract, especially in the urinary bladder. Villous adenoma of the renal pelvis was first reported in 2002 by Park et al. and until 2021 there are only 14 cases reported in the English literature.[3] It has a slight predominance in male patients in the seventh decade of life, its most usual clinical presentation is hematuria, non-specific lower urinary tract irritative symptoms, and mucosuria.[2–4] In most cases, they are incidental findings with more frequent presumptive clinical diagnoses of tuberculosis or pyonephrosis.[4] Another factor that makes suspicion difficult is that most of these neoplasms are associated with hydronephrosis secondary to renal lithiasis and complicated with abscessed pyelonephritis, as occurred in our case.[2,5] The diagnosis of this entity becomes a real challenge because it is considered a diagnosis of exclusion with very non-specific symptoms and imaging findings. The pathogenesis of these neoplasms is believed to be associated with a chronic inflammatory background derived from recurrent pyelonephritis or urolithiasis, hence these neoplasms present in advanced stages of life, beginning as intestinal metaplasia with progression to an adenoma and only in a few cases to adenocarcinoma in situ or an invasive adenocarcinoma.[1–3,5]

Grossly, villous adenoma is described as an exophytic neoplasm, with a villous or papillary appearance, indistinguishable from the papillary urothelial neoplasm more commonly found in the urinary bladder during cystoscopy. Histologically, it is identical to the villous adenoma of the colon, consisting of filiform projections lined by pseudostratified columnar epithelium with abundant cytoplasm containing mucin as well as interspersed goblet cells and low or high-grade

atypia.[2,3] The adjacent urothelial mucosa frequently shows cystitis/pyelitis glandularis and intestinal or squamous metaplasia. Immunohistochemically, positivity for CEA, CK20, CDX2, and SATB2 is consistent in most cases, and it is essential to rule out through imaging studies that it is not a distant metastasis, particularly from the digestive tract, which was also considered in our case.[2,3] It has been described that a subgroup of villous adenomas of the urinary bladder and urachus are associated with APC, PRKDC, ROS1, ATM, and KMT2D mutations.[2]

Currently, there is no consensus for the management and follow-up of patients, however, the long-term prognosis is excellent if the surgical resection is complete, even though in our case the ureteral surgical border presented intestinal metaplasia, after a one-year follow-up there is no evidence of recurrence or distant disease.[2,3]

4. Conclusion

The clinicopathological diagnosis of villous adenoma in the renal pelvis is a real challenge for the multidisciplinary team. As it is a diagnosis of exclusion, once clinical mimics have been ruled out, it should be considered a remote possibility. Despite not currently having standardized protocols for the management of these patients, total resection of the neoplasia and its thorough histopathological examination are extremely important, given the association with adenocarcinoma and consequently its prognostic impact.

Declaration of competing interest

None.

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