Case Report

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High-Grade Urothelial Carcinoma in a Pelvic Ectopic Kidney: A Case Report

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ABSTRACT

Background: The renal ectopy is a rare clinical condition that occurs due to a defect in the embryogenesis of the kidneys, causing them to occupy different locations than usual. The urothelial carcinoma of the upper urinary tract in an ectopic kidney represents a rare clinical entity with few cases described in the literature.

Case Presentation: We report here the case of a 70-year-old patient, ex-smoker, diabetic and hypertensive, who presented a high-grade urothelial carcinoma in the right pelvic kidney, whose symptoms were macroscopic hematuria and hypogastric pain. The report was prepared from information held in the Medical Archives and Statistics Department of the Urology Department of the Universitary Hospital of the Federal University of Maranhão in São Luís, Maranhão, Brazil. The patient had risk factors and a typical course of the disease. He was examined according to the diagnostic algorithm recommended in the literature and later subjected to the procedure of total nephrouretectomy with laparoscopic removal of the bladder cuff. He is being followed up on an outpatient basis, with a 6-month follow-up free of neoplastic disease.

Conclusion: Despite anatomical differences, the therapy is still similar to that of a neoplasm in an orthotopic kidney, provided a thorough anatomical examination is performed prior to surgery. The laparoscopic approach can optimize surgical treatment and shorten hospital stays when performed by an experienced surgeon.

Keywords: Upper Tract Urothelial Carcinoma; Ectopic Kidney; Pelvic Kidney; Renal Cell Carcinoma; Case Report

Background

The renal ectopy is a rare clinical condition that occurs due to a defect in the embryogenesis of the kidneys, causing them to occupy different locations than usual. Ectopic kidneys can be classified according to their location, and may have a pelvic, abdominal, thoracic, iliac, contralateral or cross-sectional topography [1]. Among these locations, the pelvic kidney occurs due to an irregularity in the ascension of the organ during the sixth to eighth week of embryonic life and may be associated with other malformations, such as caliectasis, hydronephrosis and nephrolithiasis [2]. On the other hand, the urothelial carcinoma is the most common bladder tumor and the fourth most common histological type in renal neoplasms [3,4]. About 10% of urothelial tumors are located in the upper urinary tract [5]. The incidence of an urothelial tumor in ectopic kidneys like its main clinical manifestation and macroscopic hematuria is rare. The examination of the upper urinary tract occurs through imaging exams, such as ultrasound, computed tomography and magnetic resonance imaging. Retrograde access to ureterorenoscopy is usually not feasible in pelvic kidneys, due to distortions of the ureteral anatomy. The accurate diagnosis of the urothelial tumor of the upper urinary tract in the pelvic kidney by means of imaging tests enabled good surgical planning. Despite kidney ectopia and vascular variations, a total nephroureterectomy with resection of the bladder cuff can be performed safely and laparoscopically, which enables a good oncological result.

Case Presentation

A 70-year-old man was admitted to the Urology outpatient clinic of the Universitary Hospital of the Federal University of Maranhão (HUUFMA) with a complaint of macroscopic hematuria, associated with severe hypogastric pain, with a three-month development period. He denied trauma to the region or other associated symptoms. He had diabetes mellitus and systemic arterial hypertension as comorbidities. He was a smoker, but had stopped smoking 40 years ago (smoking load 10 packs / year). No palpable abdominal masses were observed during the physical examination. The laboratory investigation revealed hypochromic microcytic anemia and impaired renal function (serum urea 68; creatinine clearance 32.3 ml / min). A simple urinalysis revealed hematuria and leukocyturia. An Urinary cytology was suspected for high-grade urothelial carcinoma. An urethrocystoscopy did not reveal bladder injuries. An urotomography (Uro-CT) showed an ectopic right kidney, positioned in the midline, anterior to the iliac vessels, with a hypodense exophytic image in the lower cortical region, measuring 2.5 cm in its largest diameter, without impregnation with the contrast medium, characterizing a simple cyst. The late excretory phase of contrast medium had a filling failure in the lower third of the kidney, suggesting the possibility of neoplasia. The left kidney was topical with several hypodense, rounded lesions of various sizes, the largest being 4.6 cm in the middle third and 3.2 cm in the upper exophytic third, without being impregnated with the contrast agent.

The radiological examination of the urinary tract was supplemented by an uro-resonance, which identified an area of irregular urothelial thickening in the right kidney involving the lower calyx group and the renal pelvis, with heterogeneous contrast enhancement, suggesting an urothelial tumor. The left common iliac artery was in close contact with the upper border of the right kidney and there was no sign of renal vascular invasion. The patient underwent a nephroureterectomy and removal of the bladder cuff via a laparoscopic approach. During the procedure, a polar renal artery exiting the common iliac artery and a large renal artery exiting the distal aorta were identified. The patient presented a favorable postoperative recovery and was discharged in good clinical condition on the fifth postoperative day. The histopathological study of the surgical specimen confirmed a high-grade urothelial carcinoma of the renal pelvis and infiltration of the kidney into perirenal adipose tissue (pT4), an angiolymphatic neoplastic invasion was also found. The hilum and renal sinus were free of neoplastic involvement, as were surgical margins. During the clinical follow-up examination, after 3 months of surgery, the patient presented a new episode of macroscopic hematuria. The diagnostic examination identified bladder injuries on urethrocystoscopy. The patient underwent a transurethral resection of the bladder with histopathological findings of a non-invasive low-grade urothelial carcinoma. After 6 months, he was free from neoplastic diseases.

Discussion

During the development of the kidney embryo, the organs reach maturity in the sacral area. During organogenesis, around the sixth to eighth week of life, the kidneys ascend into the retroperitoneal region [6]. Failures during this process result in ectopic kidneys, with the pelvic topography being the most common [7]. Anatomical changes are often associated with poor organ rotation and slightly predominate to the left [8]. In most cases, the patient is asymptomatic and an ectopia is discovered during imaging tests [9]. In turn, the urothelial carcinoma of the upper urinary tract is another rare clinical disease and represents 5 to 10% of the neoplasms of the urinary system that are often associated with other urothelial neoplasms [5]. It has a higher incidence in men between the ages of 60 and 70 and a strong association with smoking. The most common clinical presentation is macroscopic hematuria [5]. Halalsheh, et al. [10] reported the case of a patient with flank pain and hematuria whose examination identified a calcinal tumor in a poorly rotated right kidney who was treated with a radical nephroureterectomy with removal of the bladder cuff. The histopathological study showed a low-grade urothelial carcinoma [10]. Philipraj, et al. [11] described a renal mass in a pelvic kidney that exhibited suprapubic pain and difficulty urinating. A kidney tumor suspected of being a renal cell carcinoma was detected by computed tomography. Radical nephroureterectomy was performed and revealed a grade II transitional cell carcinoma [11].

Arisawa, et al. [12] reported a finding of papillary tumor of the renal pelvis during pyelolithotomy performed for the treatment of choriform calculus in a pelvic kidney. After a pathological diagnosis of freezing of the renal pelvis lesion, which proved to be a low-grade transitional cell carcinoma, the procedure was complemented with a nephroureterectomy with resection of the bladder cuff [12]. In 1984, Terai, et al. [13] described a transitional cell carcinoma in a pelvic kidney, two years after resection of a bronchogenic carcinoma in the right lung. The patient complained of abdominal pain and had pelvic mass [13]. The investigation of urothelial carcinoma in patients with ectopic kidneys follows the same principles of propedeutics in patients without known anatomical abnormalities. The only limitation is due to the tortuosity of the ureter that makes the retrograde endoscopic examination of the upper urinary tract in pelvic kidneys unfeasible. CT urography has a sensitivity of 67% to 100% and a specificity of 93% to 99%, which is regarded as the most precise image examination [14]. A urography-resonance is preferably indicated for patients with contraindications to radiation and / or iodinated contrast and in the suspicion of vascular invasion by tumor; even in this scenario, CT urography remains the preferred imaging method for the diagnosis and delimitation of the disease [15]. A comprehensive and complete preoperative evaluation is recommended for these patients, particularly due to the anatomovascular malformations associated with the disease, such as multiple renal arteries or renal arteries emerging from different vessels of the aorta.

Vascular studies may also be indicated for better surgical planning [16]. Identifying the proximity of the vessels to the kidney mass is necessary to avoid intraoperative complications such as bleeding. The principles of urothelial cancer treatment take into account the histological grade, number, size and location of the lesion, the patient's comorbidities and the surgeon's experience. Whenever possible, total nephroureterectomy with removal of the bladder cuff is the procedure of choice, which can be performed using the open or laparoscopic approach [5]. A multicentric study conducted in France suggested that the laparoscopic approach to large tumors would be associated with metastatic spread in some patients [16]. However, randomized studies have shown similar cancer outcomes regardless of the approach [5]. Despite the controversy surrounding the issue, the laparoscopic route is still preferred by some surgeons because it is less invasive and involves shorter hospital stays [17]. Adjuvant or neoadjuvant chemotherapy in the treatment of this disease still has controversial indication, mainly due to the comorbidities of patients and the loss of renal function after radical surgery. Radiotherapy is not effective, whether used alone or associated with chemotherapy [5]. The prognosis is worse, the higher the extent of tissue invasion and the life expectancy in 10 years for pT4 tumors <10% [5]. The advanced age at the time of surgery, the presence of multifocal tumors and lymphatic invasions indicate greater disease severity.

Postoperative follow-up care is essential and requires careful examination for local recurrences, distant metastases, or the presence of other urothelial cancers, especially a bladder tumor. The main site of recurrence is the bladder and, to a lesser extent, the remaining kidney. The relationship between ectopic kidney and renal neoplasia is still unclear in the literature, but there does not seem to be an increased risk of malignancy in connection with renal ectopia [10]. However, this relationship can present an uncommon challenge for urologists, especially with regard to the surgical approach (Figures 1 & 2).

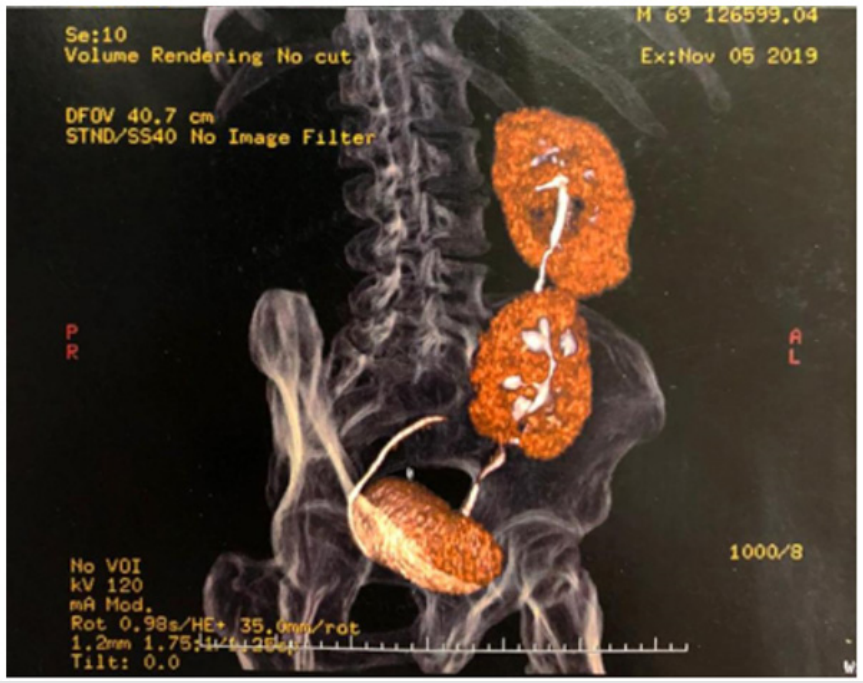

Figure 1: Computed tomography of the abdomen with three-dimensional reconstruction, showing left kidney with topical location and right kidney with low location, in the midline anterior to the iliac vessels, with the pelvis directed anteriorly.

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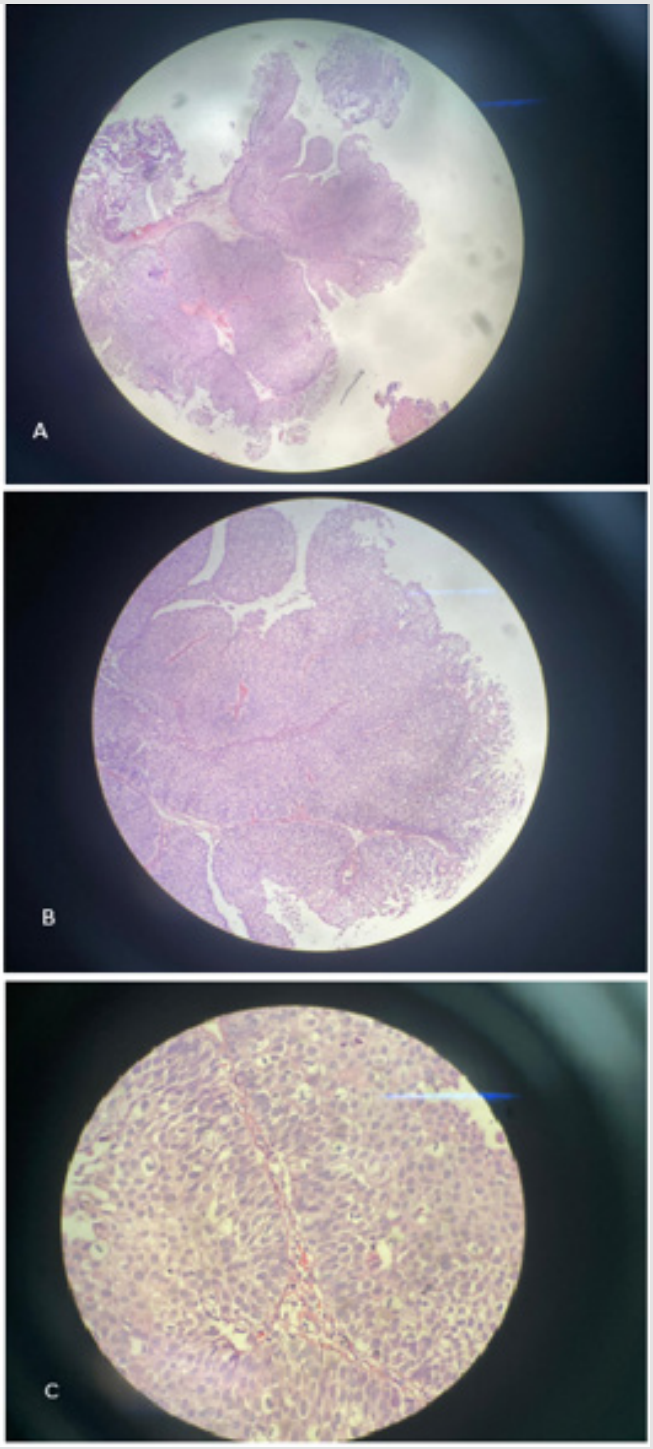

Figure 2: Cytoarchitecture compatible with high-grade urothelial carcinoma of the renal pelvis, with angiolymphatic invasion and free surgical margins.

a) Small magnification image showing lesion of papillary architecture, with discrete cellular pleomorphism and vascular axis responsible for tumor nutrition.

b) Medium magnification image.

c) In the high magnification image, cell atypia stands out, with the presence of a cytopathic effect and cytoplasm with a whitish halo.

Conclusion

Urothelial carcinoma of the upper urinary tract in an ectopic kidney is a rare disease of which only a few cases have been reported in the literature [17]. Despite anatomical differences, the regimen of therapy is still similar to that of a neoplasm in an orthotopic kidney, provided a thorough anatomical examination is performed prior to surgery. The laparoscopic approach can optimize surgical treatment and shorten hospital stays when performed by an experienced surgeon.

Declarations

Ethics Approval and Consent to Participate

This report was approved by the Research Ethics Committee of the Universitary Hospital of the Federal University of Maranhão, CEP/HU-UFMA, with the Certificate of Presentation for Ethical Assessment 39411820.0.0000.5086.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

Availability of Data and Materials

Not applicable.

Competing Interests

There are no authors' conflicts of interest.

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