Case Report

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Urothelial Carcinoma of the Renal Pelvis with Synchronous Ipsilateral Collecting Duct Carcinoma: Two Case Reports

동측 신장에서 발생한 동시성 집합관세포암종과 요로상피세포암: 2예 보고

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Synchronous renal malignancies are seldom encountered or diagnosed post-renal resection. A combination of renal cell carcinoma (RCC) and urothelial carcinoma (UC) is most commonly reported. Typically, the RCC subtype is clear-cell RCC; however, a combination of collecting duct carcinoma (CDC) and UC has rarely been reported in the existing literature. Here, we present two cases of synchronous renal malignancy, specifically a combination of CDC and UC, in the ipsilateral kidney.

Index terms Kidney Neoplasms; Collecting Duct Carcinoma; Urothelial Carcinoma; Synchronous Tumor

INTRODUCTION

Because the synchronous occurrence of multiple primary cancers in a single organ is rare, two or more different types of cancers in the ipsilateral kidney are rarely encountered. Most synchronous renal malignancies are combinations of renal cell carcinoma (RCC) and urothelial carcinoma (UC). In most cases, the RCC subtype is clear cell RCC. However, the combination of UC and collecting duct carcinoma (CDC) in the same kidney is extremely rare and has been reported in only a few cases. The radiological findings in these cases are variable and

can be challenging to distinguish from those of other renal tumors or even metastatic diseases. Herein, we present two cases of synchronous UC and CDC in the same kidney and discuss the radiological findings and diagnostic challenges associated with these rare multiple primary tumors.

CASE REPORTS

CASE 1

A 77-year-old male was referred to our institution for the evaluation of an incidentally detected mass in the renal pelvis. The lesion was detected when the patient underwent contrast-enhanced abdominopelvic CT after spinal surgery at another hospital. During his visit, he experienced lower back pain, but no other symptoms. The patient had a history of distal gastrectomy for a benign gastric ulcer. Initial laboratory findings revealed increased blood urea nitrogen and creatinine levels of 33 mL/dL (normal range: 8–26 mL/dL) and 1.3 mL/dL (normal range: 0.8–1.2 mL/dL), respectively. Modification of Diet in Renal Disease study estimated that glomerular function was decreased at 56.9 mL/min/1.73 m^2 (normal range: 60–160 mL/min/1.73 m^2); however, urine analysis revealed no abnormal results, including urine blood or white blood cell (WBC) counts.

On initial urography CT, an approximately 3.7 cm lobulated mass was detected in the right renal pelvis (Fig. 1A). The lesion had invaded the renal sinus fat and parenchyma (Fig. 1B, C). The lesion measured as isodense at 34 Hounsfield units (HU) in the pre-contrast phase, exhibited heterogeneous enhancement (100 HU) during the nephrographic phase, and showed washout (78 HU) in the excretory phase. UC components were detected using urine cytology.

With a tentative diagnosis of UC in the right renal pelvis, a right laparoscopic nephroureterectomy was performed. Histopathological examination revealed two different tumor types: CDC without lymphovascular invasion (pT1a) and papillary UC with subepithelial connective tissue invasion (pT1) without lymph node metastasis (Fig. 1D). A three-month follow-up CT scan after resection revealed multiple pulmonary and bone metastases (Fig. 1E, F).

CASE 2

A 48-year-old male was referred to our hospital with a left renal mass detected by abdominal CT at another hospital. The patient had left flank pain for a few weeks without any other symptoms such as hematuria. No specific family or medical history was recorded. An initial laboratory test revealed a slightly increased WBC count of 14990/μL (normal range: 3500–10000/μL) with no other abnormal results.

Initial abdominal CT with contrast enhancement revealed a 1.3 cm sized papillary mass with mild enhancement in the left upper calyx (Fig. 2A). Another 3 cm ill-defined mass with mild enhancement was detected in the medullary region of the left kidney (Fig. 2B). The mass was isodense (28 HU) compared to the renal cortex in the pre-contrast phase, homogeneously enhanced (58 HU) in the nephrographic phase, and progressively enhanced (74 HU) in the excretory phase. During the nephrographic phase, an enhanced filling defect was observed in the upper calyx of the left kidney. Multiple enlarged lymph nodes with necrotic portions were observed in the left renal hilar and para-aortic regions (Fig. 2C).

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Fig. 1. A 77-year-old male with synchronous collecting duct carcinoma (pT1a) and papillary urothelial carcinoma (pT1) of the right kidney.
A. Initial CT urography reveals a lobulated mass with mild enhancement (arrows) in the right renal pelvis at the nephrographic phase. At the excretory phase, this lesion is shown as a lobulated contrast-filling defect. The mean density of the mass is 34 HU, 100 HU, and 78 HU in the pre-contrast, nephrographic, and excretory phases, respectively.
B. The mass located at the right renal pelvis shows invasion of the renal sinus fat and renal parenchyma (dashed arrows). Papillary mass in right renal pelvis is also noted (arrows).
C. Coronal CT image of the excretory phase shows the renal mass located at the right renal pelvis (arrow) with the renal sinus fat and renal parenchyma invasion (dashed arrow).
D. The resected gross specimen shows a grayish-white granular and papillary mass (arrow) in the renal pelvis. The tumor was composed of urothelial carcinoma in histological examination. Another grayish lobulated mass (dashed arrow) is located at the right kidney medullary region abutting the papillary mass. The lesion was confirmed as collecting duct carcinoma, histologically. In immunohistochemical examination, both tumors showed positive staining at CK and HMW-CK. However, CK20 staining was only positive in urothelial carcinoma (not shown).
CK = cytokeratin, HMW-CK = high molecular weight cytokeratin, HU = Hounsfield units

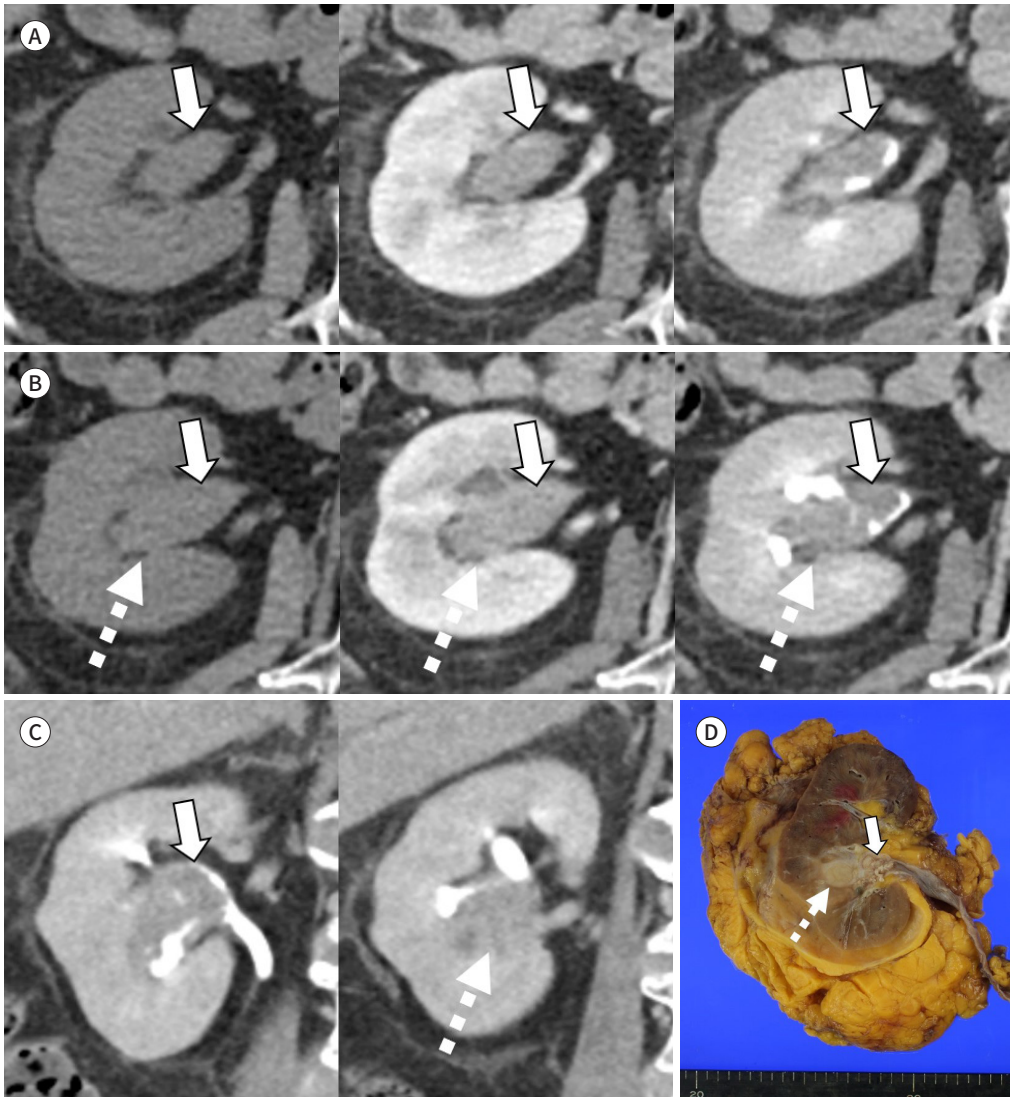


Fig. 1. A 77-year-old male with synchronous collecting duct carcinoma (pT1a) and papillary urothelial carcinoma (pT1) of the right kidney.
E, F. Chest CT with lung window setting (E) and PET-CT (F) show lung and pelvic bone metastases (arrows) three months after surgery.

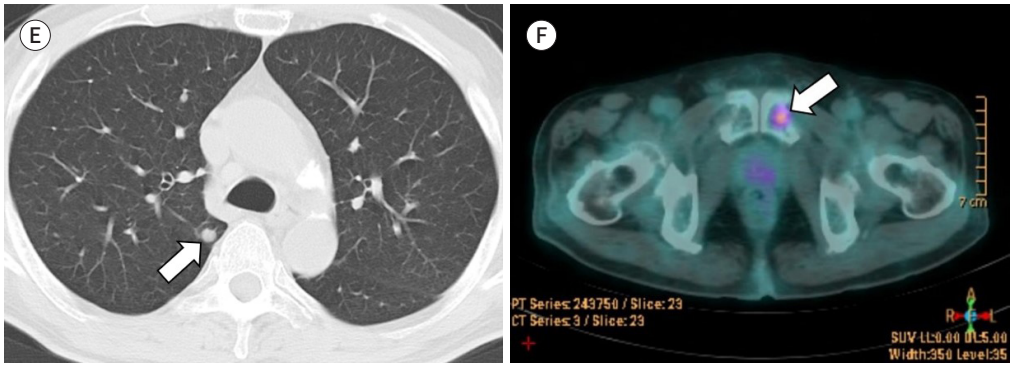
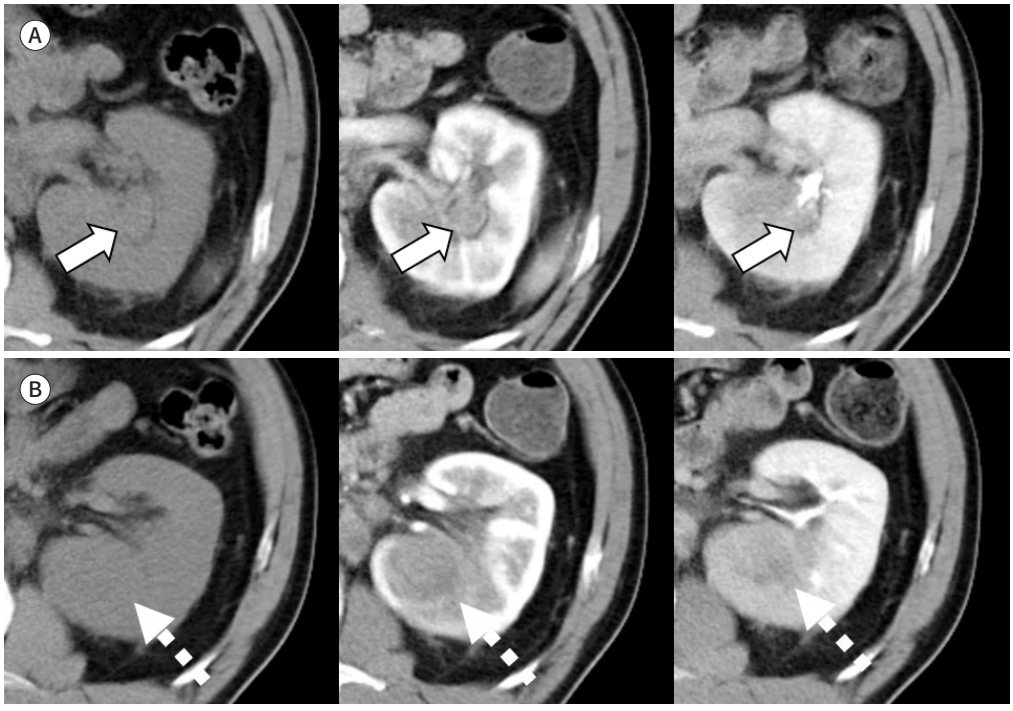


Fig. 2. A 48-year-old male with synchronous papillary urothelial carcinoma and collecting duct carcinoma at the left kidney.
A. Initial abdominal CT reveals a 1 cm sized papillary lesion (arrows) in the left kidney upper calyx. This lesion shows mild contrast enhancement.
B. Another 3 cm-sized mass with an ill-defined margin (dashed arrows) is located at the medullary region of the left kidney. This lesion shows progressive enhancement. The mean density of the mass is 28 HU, 58 HU, and 74 HU in the pre-contrast, nephrographic, and excretory phases, respectively.
HU = Hounsfield units



With a tentative diagnosis of UC with calyceal and renal parenchymal invasion and lymph node metastasis, left radical nephrectomy was performed. A gross pathological examination revealed an intraluminal papillary mass in the upper calyx (Fig. 2D). The mass was diagnosed as papillary UC without subepithelial connective tissue invasion (pTa). Another mass was

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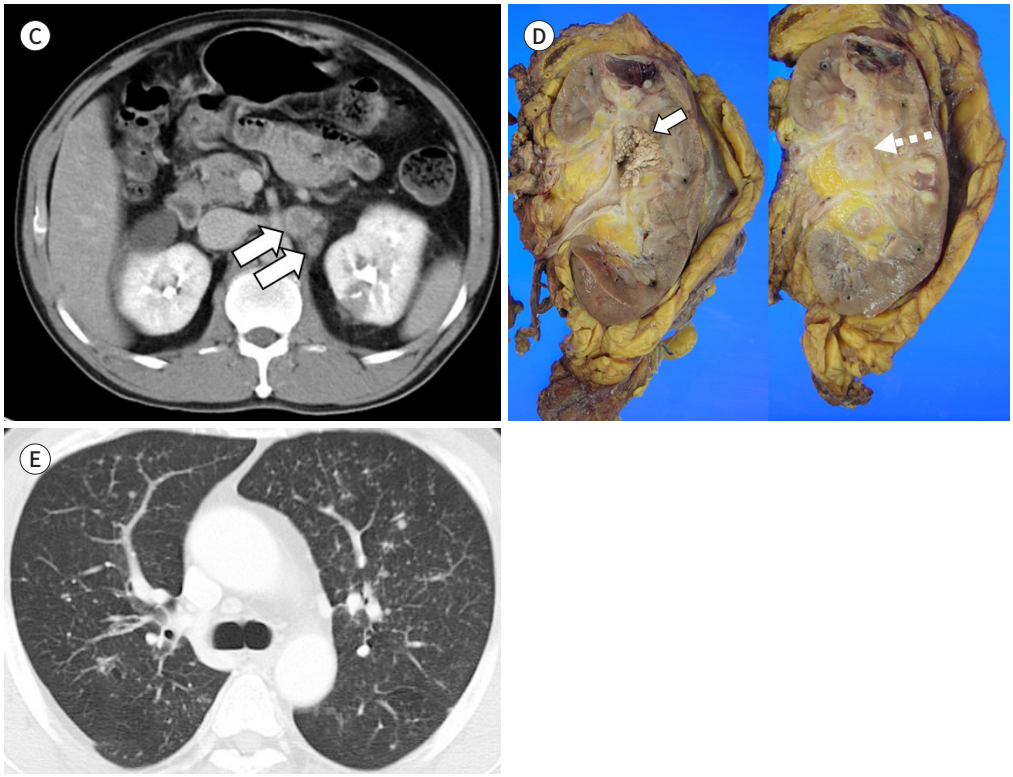
Fig. 2. A 48-year-old male with synchronous papillary urothelial carcinoma and collecting duct carcinoma at the left kidney.
C. Contrast-enhanced CT shows multiple enlarged lymph nodes (arrows) with a central low density in the left renal hilar and para-aortic regions.
D. Gross section of the kidney shows a grayish papillary mass (arrow) in the upper calyx. In histological examination, the tumor was composed of urothelial carcinoma. Another white lobulated mass (dashed arrow) is located at the left kidney medullary region. The mass was confirmed as collecting duct carcinoma, histologically. The former lesion showed negative staining in high-molecular-weight cytokeratin and vimentin. However, the latter portion showed positive staining in high-molecular-weight cytokeratin and vimentin (not shown).
E. Two weeks after resection, chest CT with lung window setting shows multiple nodules in both lungs, compatible with pulmonary metastases.



identified in the renal medullary region, demonstrating invasion into the renal sinus fat (Fig. 2D). Pathological staining (high molecular weight cytokeratin and vimentin) confirmed the diagnosis of CDC (pT3a). Metastases of CDC were observed in the regional lymph nodes. The left adrenal gland exhibited hemorrhagic changes without any masses.

After two weeks, the patient underwent chest and abdominal CT with contrast enhancement for staging and further chemotherapy. Chest CT revealed multiple nodules in both lungs (Fig. 2E) and multiple heterogeneously enhanced lymph nodes in the left upper paratracheal, subcarinal, hilar, right middle lobar, interlobar, and segmental areas. Additionally, multiple low-density masses were detected in both liver lobes on the abdominal CT. Subsequently, FDG PET/CT revealed multiple bony metastases in the thoracolumbar spine. The multiple hepatic masses and bony metastases were believed to be metastatic lesions originating from CDC. The patient underwent chemotherapy with bone radiation therapy for one month and was lost to follow-up after discharge.

This case report conformed to the ethical guidelines of the 1975 Declaration of Helsinki.

And we received informed consent from each patient for the study.

DISCUSSION

Synchronous primary neoplasms of the same kidney are rare and only a few cases have been reported. The most frequently reported type is the combination of RCC and UC. However, the combination of CDC and UC is extremely rare and has only been reported in a few cases (1-3).

CDC was first reported by Jimenez et al. and was listed as a major subtype of RCC in the 2002 WHO classification. CDC, also known as Bellini carcinoma, is a rare variant of RCC that accounts for less than 1% of all renal cancers (4). CDC shows a highly aggressive biological behavior and poor prognosis, with frequent lymph nodes and distant metastases. At initial diagnosis, 40% of patients present with distant metastases such as hepatic, pulmonary, or adrenal metastases (5). The CDC originates in the distal part of the collecting duct. The tumor stroma showed increased fibrous tissue hyperplasia and collagen expression (6). Several studies have described radiological findings in CDC. Owing to its origin, the tumor center is located at the renal medulla, infiltrating the cortex and renal pelvis. Usually, irregular borders and expansile and infiltrative growth patterns are observed at the center. On preoperative imaging, renal medullary and pelvic invasion and infiltrative spread were detected in 94% and 67% of patients, respectively (7). On CT, the tumor parenchyma was denser than the surrounding normal renal tissue. On MRI, CDC exhibits isointensity on T1-weighted images and low signal intensity on T2-weighted images compared with the signal intensity of renal tissue (7). Enhancement of CDC tumors showed a hypovascular pattern in most cases. Another difference between CDC and other common renal cancer pathology types is the relatively high FDG uptake observed on PET/CT in CDC.

Primary pelvicalyceal UC accounts for approximately 15% of all kidney tumors, and aggressive high-grade UC can exhibit renal parenchymal invasion with an infiltrative growth pattern. UC occurs in the sixth and seventh decades of life, and male are affected three times more often than female (8). CT urography is the modality of choice for diagnosis. During the excretory phase of CT, pelvicalyceal UC appears as a filling defect in the renal pelvis, a mass, or circumferential thickening of the urinary tract (9). Additionally, the nephrographic or corticomedullary phase can help detect the heterogeneous enhancement of the mass and invasion of the renal sinus fat and parenchyma. However, large pelvicalyceal UC with renal invasion can mimic other renal diseases, such as RCC, lymphoma, or acute pyelonephritis with abscesses. In some cases, MR urography can aid in the differential diagnosis of renal UC and in tumor staging using diffusion-weighted imaging and apparent diffusion coefficient maps (10).

Various theories have been suggested regarding synchronous tumor occurrence in the kidney: 1) two different cell lines proliferate simultaneously, resulting in two phenotypically different tumors, 2) tumors arising from common precursor stem cells result in two unrelated neoplasms, and 3) two different isolated tumors occur incidentally at the same anatomic site (10). In each of our cases, two different tumors were found at the same anatomical location, with margins in two cases, and two types of tumor cells in an infiltrative mass without mar-

gins in one case. The first two theories may apply to our case because of the location of the two different tumors. However, the mechanism and relationship between simultaneous primary renal tumors remain unclear.

Although the incidence is low, ipsilateral synchronous RCC and UC, including their radiological findings, have been reported in the literature. However, to the best of our knowledge, only two cases of synchronous CDC and UC in the same kidney have been previously reported (1-3). The radiological findings in these cases are variable and can be challenging to distinguish from those of other renal tumors or even metastatic diseases.

In a retrospective review of the radiological findings in our cases, the presence of synchronous cancers was not recognized using only the initial CT and US findings. In both cases, the lesions were heterogeneous with enhanced infiltrative masses in the renal pelvis and renal parenchymal invasion. Although the initial radiologic diagnosis of the two patients was UC with metastatic lymphadenopathy and distant metastasis, the pathologic T stages of UC were T1 and Ta, respectively. The discrepancy between the initial diagnosis and pathological results may be due to the aggressive nature of synchronous CDC.

Owing to the aggressive behavior and high distant metastasis rate of CDC at initial diagnosis, there is a significant difference in the prognosis and treatment plan compared with other malignant renal tumors. Radiologists play an important role in the differential diagnosis of renal tumors, helping to establish treatment plans and leading to better prognoses. If initial radiological studies reveal an infiltrative renal mass or renal pelvic tumor with an infiltrative growth pattern, CDC should be considered as a tentative diagnosis.

In conclusion, we reported two rare cases of synchronous CDC and UC of the ipsilateral kidneys. Differential diagnosis of this rare renal mass is difficult, even for experienced radiologists. However, in cases of renal masses with infiltrative growth, medullary location, involvement of the renal sinus, weak contrast enhancement, and aggressive behavior with distant metastasis, it is necessary to consider not only conventional RCC and UC but also the rare combination of CDC and UC. Better treatment strategies and prognoses can be expected by determining the probabilities of the differential diagnoses.

Author Contributions

Conceptualization, all authors; data curation, all authors; investigation, B.S.B., Y.S.K.; project administration, Y.S.K.; supervision, Y.S.K.; validation, Y.S.K.; writing—original draft, B.S.B.; and writing—review & editing, Y.S.K.

Conflicts of Interest

Seong Kuk Yoon has been a Section Editor of the Journal of the Korean Society of Radiology since 2017; however, he was not involved in the peer reviewer selection, evaluation, or decision process of this article. Otherwise, no other potential conflicts of interest relevant to this article were reported. All remaining authors have declared no conflicts of interest.

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동측 신장에서 발생한 동시성 집합관세포암종과 요로상피세포암: 2예 보고

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신장의 동시성 악성종양은 드물게 발견되며, 신장 수술 후 진단된다. 신세포암과 요로상피세포암이 함께 동반되는 경우가 가장 많이 보고 되었다. 대부분의 경우 신세포암의 아형은 투명세포형 신세포암이지만, 동시성 집합관세포암종과 요로상피세포암이 동반되는 경우에 대한 보고는 매우 드물다. 동측 신장에서 발생한 동시성 집합관세포함종과 요로상피세포암의 2건의 증례를 보고한다.

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