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A CASE OF PAPILLARY RENAL CELL CARCINOMA SUGGESTIVE OF BELLINI DUCT ORIGIN

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A case of Bellini duct carcinoma is reported. A left renal tumor was detected by abdominal computerized tomography in a 76-year-old male, although he had no symptoms, such as hematuria, weight loss or flank pain. Radical nephrectomy was performed under the diagnosis of renal cell carcinoma in the left kidney.

Macroscopic examination of the resected kidney revealed a tumor 2.0 cm in diameter, with a yellow-brown cut surface, located in the renal medulla. Histological examinations showed malignant tumor cells with eosinophilic cytoplasm with a papillary growth pattern. Immunohistostaining examinations using Lectin and two kinds of monoclonal antibodies demonstrated no significant staining with soybean agglutinin, peanut agglutinin, Dolichos biflorus agglutinin, Lotus tetragonolobus agglutinin or cytokeratin, and negative staining with Tamm-Horsfall protein. Although the results of immunohistostaining did not provide support, both macroscopic and microscopic findings strongly suggested that this tumor originated from Bellini duct epithelium (Bellini duct carcinoma). The patient is alive with no evidence of disease 1 year after surgery.

Bellini duct carcinoma is a rare malignant condition and the prognosis is usually poor. Differential diagnosis from other renal or pelvic tumors is difficult and long-term careful follow-up is necessary.

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Key words: Bellini duct, Carcinoma, Kidney

INTRODUCTION

Primary epithelial tumors of the kidney are generally recognized as either of transitional cell origin arising from the renal pelvis or as renal cell carcinoma arising from the proximal convoluted tubule.

Recently, Cromie et al. described a kidney tumor with cellular elements of both renal and transitional cells and suggested that its origin was the epithelium of the collecting ducts of Bellini¹⁾. This tumor is an exclusively malignant condition.

The present case shows features suggesting that it originated from the cells of the collecting duct epithelium (Bellini duct carcinoma).

CASE REPORT

A 76-year-old man visited our hospital because an abdominal computerized tomograph (CT) taken by a local physician showed a tumor in the left kidney. There was no evidence of macroscopic hematuria, flank pain or weight loss. Laboratory examination including cytologic study of urine did not reveal any abnormalities. Chest X-rays did not show any metastases. Excretory urography showed a deformity of the left renal pelvis. Abdominal CT showed an isodense mass located near the renal pelvis of the left kidney, but there was no enhancement by contrast material (Fig. 1). Retrograde pyelography showed compression of the renal pelvis by the

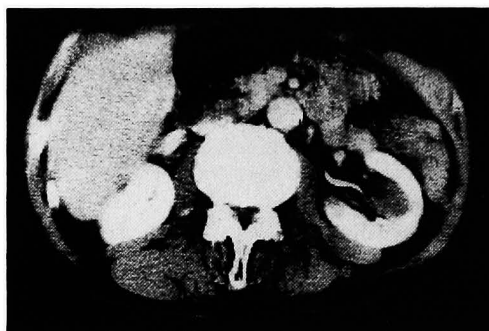


Fig. 1. Abdominal CT demonstrated an isodense mass located near the renal pelvis of the left kidney. The tumor was not enhanced by contrast media.

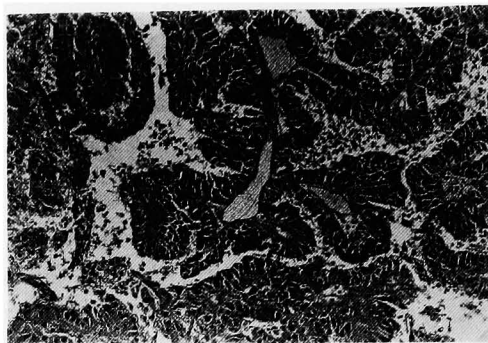


Fig. 3. Histological findings of the tumor. Malignant tumor cells with cuboidal, eosinophilic cytoplasm, nuclear polymorphism grew in a papillary pattern (H & E stain $\times 200$).

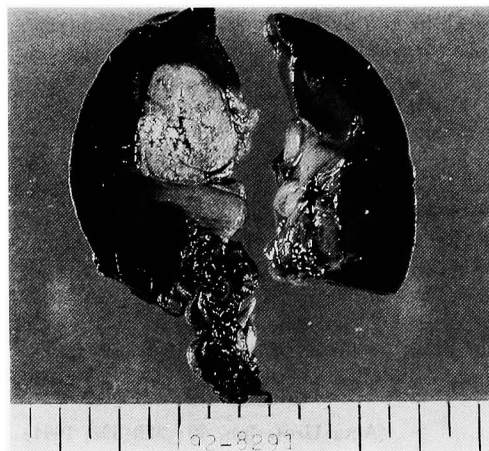


Fig. 2. Macroscopic findings of the resected kidney. A small white-yellow-brown tumor (2.0 cm in diameter) was located in the middle part of the renal medulla.

tumor (data not shown). Left radical nephrectomy was performed under the diagnosis of renal cell carcinoma. The tumor did not invade the surrounding tissue and swelling was not detected in the regional lymph node. Macroscopically, the resected kidney showed a yellow-brown tumor mea-

suring 2.0 cm in diameter, which had a white capsule and was located in the middle part of the renal medulla (Fig. 2). Histological examination showed malignant tumor cells with cuboidal eosinophilic cytoplasm, nuclear polymorphism, and distinct nucleoli. The cells were similar to the distal collecting duct epithelium and grew in a papillary pattern (Fig. 3). In some areas, tumor cells invaded the renal parenchyma beyond the capsule. Lectin and monoclonal antibodies were used in immunohistostaining and the results are shown in the table (Table 1). Immunohistochemical analysis on paraffin-embedded sections was performed with the PAP methods of Kikuchi¹¹⁾. Soybean agglutinin (SBA), peanut agglutinin (PNA), *Dolichos biflorus* agglutinin (DBA), *Lotus tetragonolobus* agglutinin (LTA) and monoclonal antibodies for cytokeratin (CK) stained slightly with tumor cells, whereas THP did not (data not shown). Electron microscopic examination was not performed. Although the results of immuno-

Table 1. Antibodies, their dilutions, and the results of immunostaining

Markers	Sources	Dilutions	Results	Results
<i>Lotus tetragonolobus</i> agglutinin (LTA)	E. Y. Lab	$\times 20$		+
Peanut agglutinin (PNA)	E. Y. Lab	$\times 100$		+
<i>Dolichos biflorus</i> agglutinin (DBA)	E. Y. Lab	$\times 100$		+
Soybean agglutinin (SBA)	E. Y. Lab	$\times 100$		+
Cytokeratin PCK1 (CK)	Lab. systems	$\times 100$		+
Tamm-Horstall Protein (THP)	Cedarlane	$\times 2000$		-

+ ; weakly positive staining, - ; negative staining

staining in our case were not clear, the tumor was strongly suspected of being Bellini duct carcinoma by macroscopic and microscopic findings. The patient is alive with no evidence of disease 1 year after surgery.

DISCUSSION

Bellini duct carcinoma is a rare exclusively malignant condition. Only 16 cases have been reported in the English and Japanese literature, since Cromie et al. reported the first case in 1979¹⁻¹⁰. Recently, tumors originating from the renal distal collecting tubules were divided into two types. The "mixed-type" is gray-white macroscopically, and histologically contains renal cell carcinoma and transitional cell carcinoma^{1,3,12}. The "papillary type"^{2,5-7,9,12} is yellow or brown-red macroscopically, with some small, pale yellow areas, and histologically, tumor cells demonstrated eosinophilic cytoplasm, nuclear polymorphism, distinct nucleoli, and are similar to distal collecting duct epithelium with a papillary growth pattern accompanied by an invasion of macrophage and fibrous connective tissue^{2,12,13}. Such tumors develop from the renal medulla and grow to reach the cortical tissue. Therefore, if the tumor is small, it would be located in the renal medulla only. In our case, the tumor was located in the medulla tissue of kidney, had a papillary growth pattern and tumor cells demonstrated eosinophilic cytoplasm. Therefore, this tumor was diagnosed as "Bellini duct carcinoma" and subclassified as "papillary type"

Recently, several investigators demonstrated immunohistochemical analysis using various antibodies against several portions of normal renal tubules^{7,9,10}. Kikuchi et al. demonstrated differential diagnosis from common papillary renal carcinoma using 8 monoclonal antibodies¹¹. Rumpelt et al.¹⁰ demonstrated 6 cases diagnosed by immunohistochemical studies using monoclonal antibodies. However, the results of immunohistochemical analysis are not clear; the results reported by several investigators differ. Aizawa et al. reported that LTA reacted with the normal epitheli-

um of proximal tubule and Bellini duct; THP reacted with the normal epithelium of distant tubule; and SBA, PNA, DBA, CK and VM reacted with the distal portion of the tubule¹⁴. Our findings show that PNA, SBA, DBA, LTA and CK reacted weakly, but THP did not react with tumor cells. Results of immunohistostaining in our case did not confirm the results by Kikuchi. A possible reason for weak staining with lectin was that the tumor was kept in formalin solution for a long time therefore the antigen was lost. However, in our case, the tumor was small and located in the renal medulla. Therefore, this tumor is strongly suspected to be "Bellini duct carcinoma"

Usually Bellini duct carcinoma is a high grade malignancy^{3,5,7,10}. In their series, Rumpelt et al.¹⁰ demonstrated a very pronounced infiltrative pattern of tumor growth, resulting in replacement of the original renal and hilar tissue by tumorous tissue without signs of compression; they suspected the tumor cells spread via interstitial, intratubular and intravenous pathways. In our case, the tumor size was 2.0 cm in diameter, and there was no evidence of lymph node or distant metastases, but microscopically tumor cells had invaded the surrounding parenchymal tissue of the affected kidney. Although the numbers of cases is small, the prognosis of Bellini duct carcinoma is usually very poor. Therefore, long-term and careful follow-up is necessary in patients with Bellini duct carcinoma.

In our case, although electron microscopic examinations were not done, this tumor was strongly suspected to be "Bellini duct carcinoma" because the histological features were typical of Bellini duct carcinoma and the tumor was located in the medulla of the left kidney.

In conclusion, Bellini duct carcinoma is a rare tumor with a high malignant potential. This tumor should be considered in the differential diagnosis of common renal cell carcinoma or urothelial cancer, and long-term, careful follow up is necessary after radical nephrectomy.

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和文抄録

ペリニ管由来が示唆された乳頭状腺癌の1例

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後 藤 章 暢

ペリニ管癌の1例を報告した。患者は76歳男性で血尿, 体重減少, 側腹部痛などの症状を有していなかったが健康診断のため近医で施行された腹部 CT で左腎腫瘍が疑われ治療目的で当科へ紹介入院となった。腹部 CT では左腎髓質に腫瘍陰影をみとめ左腎腫瘍の診断の下に根治的左腎摘出術が施行された。肉眼的に摘除腎の髓質に剖面黄褐色で直径約 2 cm の腫瘍を認めた。病理組織学的に円柱状で好酸性胞体を有する悪性細胞が乳頭状に増殖していた。レクチンおよび2種のモノクローナル抗体を用いた検討では, Soybean agglutinin, Peanut agglutinin, Dolichos biflorus

agglutinin, Lotus tetragonolobus agglutinin および Cytokeratin は弱陽性で Tamm-Horsfall protein が陰性であった。われわれの免疫組織学的検討の結果は他の報告と必ずしも一致するものではないが, 腫瘍の肉眼的および組織学的所見は, 本腫瘍がペリニ管上皮由来であることを強く示唆した。患者は手術後1年を経たが腫瘍の再発を認めず健在である。ペリニ管癌は非常に稀な腎悪性腫瘍であり一般に予後は不良である。時として他の腎癌や腎盂上皮癌との鑑別は困難であり術後長期にわたる注意深い経過観察が必要である。

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