

BELLINI'S DUCT TUMOR ASSOCIATED WITH END STAGE RENAL DISEASE: A CASE DIAGNOSED BY LECTIN IMMUNOHISTOCHEMISTRY

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We report a hemodialysis patient with an atypical renal neoplasm. The tumor cells were arranged in a two-cell pattern similar to that in the usual excretory duct systems. The histochemical staining pattern with some lectins and monoclonal antibody corresponded to the distal nephrons of normal kidney tissue. These findings enabled us to diagnose this patient as having so-called Bellini's duct tumor.

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Key words: Bellini's duct tumor, Immunohistochemistry, End-stage renal disease

INTRODUCTION

Bellini's duct tumor is a very rare neoplasm thought to originate from the distal nephrons. Only a few cases have been reported in the literature since Cromie's first report in 1979¹⁻⁴⁾. This report is of interest because it deals with the first case of Bellini's duct tumor associated with end stage renal disease.

CASE REPORT

A 63-year-old man was admitted because of gross hematuria and right flank pain.

The patient had a long history of proteinuria dating back to his youth. He was started on maintenance hemodialysis five years before admission.

On May 27, 1989, he experienced gross hematuria with an abrupt onset of right flank pain. Abdominal ultrasonography performed the next day disclosed a mass in the upper pole of the right kidney. Cystoscopy revealed bleeding from the right uretero-vesicular orifice and a retrograde pyelogram demonstrated that the upper calices of the right kidney were distorted. A computed tomographic scan of the abdomen demonstrated a subcortical hypodense right renal mass, but there was no

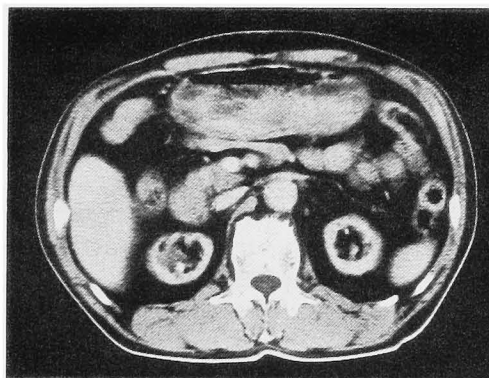


Fig. 1. Contrast-medium-enhanced CT scan showing a subcortical hypodense mass through the upper pole of the right kidney.

evidence of vascular involvement or metastases (Fig. 1). A cytological examination of the urine yielded class V cytology. These findings suggested that the renal lesion was a neoplasm arising from the renal pelvis or the medulla.

On June 13th, radical right nephrectomy was performed with the presumptive diagnosis of a renal pelvic tumor. The excised kidney was moderately atrophic. It measured approximately 7.5 by 3.5 by 3.0 cm. When sectioned, a solid and dark yellow tumor with hemorrhage was located near

the renal papilla. There was no fibrous capsule demarcating it from the surrounding renal tissue (Fig. 2). Microscopically, the tumor emerged from the acquired cyst wall and filled its lumen with papillary

growth (Fig. 3). In most parts the tumor cells were arranged as a two-cell pattern that was similar to the structure of the usual excretory duct systems. The nuclei of these cells were relatively regular in size

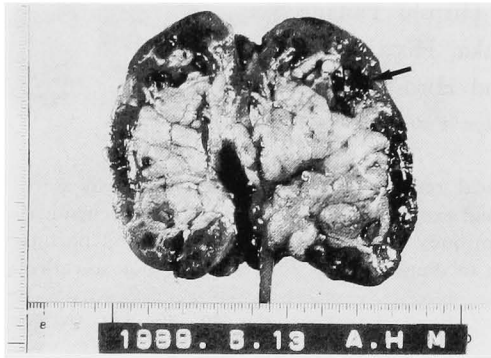


Fig. 2. Macroscopic appearance showing atrophy of the renal cortex and the mass (arrows) localized near the renal papilla.

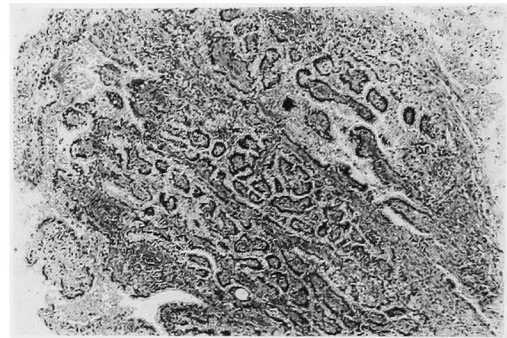


Fig. 3. Microscopic features as stained with H.E. Original magnification $\times 25$.

Table 1. Staining patterns with six lectins and one monoclonal antibody in the normal human nephrons (upper column), in the nephrons of six patients with end stage renal disease (middle), and in the nephrons adjacent to the neoplasm of our patient (lower).

Lectin/ monoclonal antibody	Segment	PCT	THL	ALH	DCT	CD			TEP	Our Case
						CO	OM	IM		
LTA		+	+/-	-	-	-	-	+/-	+/-	
		+	+/-	-	-	-	-	+/-	+/-	-
		+	+	-	-	-	+/-	+	+	
THP		-	-	+	+	-	-	-	-	
		-	+/-	+	+	-	-	-	-	-
		-	+	+	+	-	-	-	-	
PNA		-	+/-	+/-	+	+	+	+/-	+	
		-	+	+	+	+	+/-	+/-	-	+
		-	+/-	+	+	+	+	+	+/-	
SBA		-	+	+	+	+	+	+	+	
		+/-	+/-	+	+	+	+	+	+/-	+
		-	+	+	+	+	+	+	+	
DBA		-	+	+/-	+/-	+/-	+	+	+	
		-	+	+/-	+/-	+/-	+	+	+/-	+
		-	+	+	+/-	+	+	+	+	
EMA		-	+	+	+	+	+	+	+	
		-	+	+	+	+	+/-	+/-	+/-	+
		-	+	+	+	+	+	+/-	+	
UEA-1		-	-	-	-	-	-	+	+	
		-	-	-	-	-	+/-	+/-	+/-	-
		-	-	-	-	-	+/-	+	+	

and shape and exhibited little mitotic activity.

To identify the origin of this tumor in the nephron, the carbohydrate histochemistry was done at the light microscopic level using six biotinylated lectins; Lotus tetragonolobus agglutinin (LTA), Tamm-Horsfall protein (THP), peanut agglutinin (PNA), soybean agglutinin (SBA), Dolichos biflorus agglutinin (DBA) and Ulex europaeus-1 (UEA-1), and one monoclonal antibody (MoAb); Epithelial membrane antigen (EMA). The remaining nephrons in the parenchyma adjacent to this tumor and those in the renal biopsies of six dialysis patients were analyzed in this fashion as controls. Table 1 shows the staining patterns. The renal tumor reacted with lectins and MoAb specific to the lower nephrons e.g. PNA, SBA, DBA and EMA, but was not stained with LTA specific to the proximal tubules and the distal collecting ducts, THP specific to the loop of Henle or UEA-1 specific to the distal collecting duct.

These findings support the possibility that the neoplasm in our patient originates from the distal nephrons corresponding to Bellini's duct in a broad sense.

DISCUSSION

Recently, lectins have been used in immunohistochemistry to aid in glycoconjugate analysis of various lesions.

They have been utilized, in some cases, to identify the origin of a tumor, and in other cases, to observe changes of glycoconjugates in the process of a disease. However, changes in glycoconjugates that occur during the process of neoplasia or other diseases make it difficult at times to identify the cell of origin.

Some investigators have indicated that the more differentiated the renal cell carcinoma morphologically is, the greater is the lectin binding. However, some lectins (e.g. BPA, LTA and PNA) are always relatively positive independent of the differentiation of the renal tumor⁶⁾. The neoplasm of our patient was well-differentiated, and we expected to see few changes in

the lectin binding affinity.

Furthermore, in order to investigate the changes in glycoproteins of the nephron in the process of end stage renal disease, the renal parenchyma adjacent to the tumor of our patient and the renal tissues of other dialysis patients served as the controls. The staining pattern of these controls was almost equal to that of the normal kidney if the nephrons retained their original form. These results show that the effects of end stage renal disease on the lectin reactivity are negligible.

Our findings suggest that the neoplasm of this patient may have originated not from the inner medullary collecting ducts which correspond to the Bellini's duct, but from the cortical or outer medullary collecting ducts. However, some investigators maintain in a broad sense that a neoplasm arising from any segment of the distal nephrons is included in Bellini's duct tumor; therefore, we also recognized our patient's tumor as this type of tumor.

Most cases of Bellini's duct tumors reported in the past had a rapidly progressive and fatal clinical course, and these facts make it important to determine the origin of the renal tumor. However, our patient has exhibited neither recurrence nor metastases since his surgery a year ago.

The difference in the clinical course between the dialysis patients and non-dialysis patients suggests the possibility that Bellini's duct tumor belongs to another category of renal tumors when associated with the end stage renal disease just as in our case.

In conclusion, though not infallible, glycoconjugate histochemistry with lectins is the best method to establish a diagnosis of an atypical tumor arising from the distal nephrons. However, with regard to a neoplasm of dialyzed kidney different from that of non-dialyzed kidney, it remains to be proved whether or not determination of its origin has clinical significance.

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和文抄録

慢性腎不全患者に合併した Bellini 管腫瘍の 1 例

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最近われわれは、慢性腎不全のため血液透析中の患者に、腎髄質限局性の腫瘍病変を認めたが、レクチン等による組織化学染色の結果、集合管由来の腫瘍と診断されたので報告する。症例は63歳、透析歴6年の男性である。右側腹部痛および血尿を主訴に、当科を受診した。術前検査では腎盂腫瘍と診断し、根治的腎摘出術を施行した。腫瘍は腎髄質に限局、光顕的には管状、乳頭状に増殖し、two cell pattern を示していたため、集合管由来であることが疑われた。そこで、本患者の腫瘍組織並びに隣接した非腫瘍組織を各種のレクチンおよびモノクローナル抗体を用いて染色を行った。また染色パターンの比較のため、透析患者6

名の比較的構築の保たれている腎組織を同様の方法で染色した。その結果、本腫瘍組織では、PNA, SBA, DBA, EMA が陽性となり、LTA, THP, UEA-1 は陰性であった。この染色パターンは、対照とした透析腎の髄質外層集合管のパターンに一致した。以上より本腫瘍は組織化学的にも集合管由来であることが示唆された。一般に非透析腎に発生した Bellini 管腫瘍の予後はきわめて不良であるとされている。しかし自験例に再発の兆候はなく、透析腎に発生する Bellini 管腫瘍が、非透析腎のそれとは異なる、新たな疾患単位である可能性が示唆された。

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