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Mucinous cystadenoma of the pyelocaliceal system: a report of 3 examples and an analysis of 17 previously published cases ☆

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ABSTRACT

We report 3 patients all men between 45 and 64 years of age with unilocular or multilocular mucinous cystadenomas of the kidney. One tumor arose from the renal pelvis, and 2 involved the entire pyelocaliceal system. The tumors measured between 2.4 and 37 cm in greatest dimension. Two patients were asymptomatic, and 1 had recurrent attack of acute pyelonephritis. Microscopically, the morphology and immunophenotype (CK20, MUC2, and CDX2 positive) of the tumors were similar to the colonic adenomas. Two patients were asymptomatic 24 and 64 months after surgery, including the patient with mucinous cystadenoma and intramucosal carcinoma. One patient died of acute myocardial infarction, and his tumor was an autopsy finding. Only 17 cases of mucinous cystadenomas and 5 cases of mucinous cystadenocarcinomas have been reported. Of the 17 mucinous cystadenomas, 2 arose in horseshoe kidneys. The mean size of these neoplasms was 15 cm (2.4-37 cm). Despite their large size, some patients with mucinous cystadenomas were asymptomatic. Sixty percent were associated with renal lithiasis. Thirty percent progressed to mucinous adenocarcinomas, and only 2 cases showed areas of intramucosal carcinomas. Two cases were associated with carcinoid tumors, similar to those reported in the appendix. Most patients were asymptomatic after surgery, and only 1 patient died by abdominal sepsis related to adenomucinosis. The 3 examples of mucinous cystadenomas of the pyelocaliceal system reported here, and those previously published indicate that they are very uncommon neoplasms with morphology and intestinal immunophenotype similar to the colonic adenomas.

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Mucinous cystic tumors of the renal pelvis or the entire pyelocaliceal system are uncommon neoplasms. Most are mucinous cystadenomas and less frequently mucinous cystadenocarcinomas. Mucinous cystadenomas involving the renal pelvis or the entire pyelocaliceal system were first recognized more than 80 years ago [1]. Because they are exceedingly rare, little is known about their natural history, pathogenesis, morphologic, and immunohistochemical features. In fact, the publications on these neoplasms are limited to a few cases [2-7]. Moreover, these rare tumors appear to have a significant malignant potential as shown by their progression to mucinous adenocarcinoma [8-11]. Mucinous cystadenocarcinomas of the pyelocaliceal system were described more than 50 years ago, although they are less common than cystadenomas [12-16].

The purpose of this report is to describe 3 examples of mucinous cystadenomas, 1 involving the renal pelvis and the other 2 involving the entire pyelocaliceal system. In addition, we describe their natural history, morphologic features, immunohistochemical profile, and differential diagnosis. These 3 renal mucinous cystadenomas will be compared with those previously reported. A brief comment about the mucinous cystadenocarcinomas recorded by the Surveillance, Epidemiology, and End Results (SEER) program is included.

1. Materials and methods

The 3 cases of mucinous cystadenomas were retrieved from the pathology files of the Medica Sur Clinic of Mexico City, General Hospital of Mexico City, and Hospital Juan Graham Casasús from Villahermosa, Tabasco, Mexico. Multiple hematoxylin and eosin–stained sections were available for review in all 3 cases. The clinical and follow-up information was obtained from the medical charts.

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From selected paraffin blocks, additional sections were obtained for immunohistochemical analysis. The following antibodies were used: cytokeratin (CK) 7 (1:200; Biocare, Concord, CA), CK 20 (1:400; Biocare), CDX 2 (1:100; BiosB, Sta Barbra, CA), MUC2 (1:200; BiosB), Chromogranin (1:100; Biocare), synaptophysin (1:100; Biocare), CD56 (1:100; Biocare), and carcinoembryonic antigen (CEA) (1:100; Biocare).

In addition, we analyzed all cases of mucinous cystadenomas of the pyelocaliceal system reported in the English literature. The findings on mucinous cystadenocarcinomas previously reported were compared with those of the National Cancer Institute's SEER Program from 1973 to 2006.

2. Results

2.1. Clinical cases

2.1.1. Case 1

Several months before admission, a large asymptomatic abdominal mass was palpated in this 64-year-old man. A computed tomography (CT) of the abdomen showed right hydronephrosis and nephrolithiasis. His history revealed rheumatic mitral valvulopathy and several posteroseptal myocardial infarctions. He also had a history of cerebral infarction involving the temporal lobe. He was admitted to the emergency department of the General Hospital of Mexico City with intensive acute chest pain. An electrocardiogram revealed a massive myocardial infarction, which led to his death. An autopsy was performed.

2.1.2. Case 2

During a routine physical examination for hypertension at Medica Sur Clinic of Mexico City, a large and painless abdominal mass was palpated in this 54-year-old man. The large mass extended from the left upper to the lower abdominal quadrant as well as the mesogastrium and epigastrium. The patient was asymptomatic and did not lose weight. The laboratory data were unremarkable, except for a slight rise in serum CEA. A CT showed a multiloculated cystic mass involving the left kidney. The locules showed thin septae with a minor solid component and calcifications. A left nephrectomy was performed.

2.1.3. Case 3

A 45-year-old man was admitted to the Juan Graham Casasús Hospital of Villahermosa Tabasco Mexico with a history of recurrent attacks of acute pyelonephritis. A CT of the abdomen showed left hydronephrosis without neophrolithiasis. A left nephrectomy was performed.

2.2. Gross features

Grossly, 2 mucinous cystadenomas involved the entire pyelocaliceal system and the other one involved only the renal pelvis. The kidneys of the first 2 patients were markedly enlarged measuring from 20 to 37 cm in greatest dimensions. They were totally replaced by a multiloculated cystic mass involving the renal pelvis and calyces. The locules measured from 5 to 10 cm, contained abundant mucin, and had a white or pink smooth inner lining. In some areas, however, there were small nodules, granular or micropapillary structures (Figs. 1 and 2). Some of the nodules measured up to 1.8 cm in greatest dimension. In some locules, there were calculi. Only a remnant rim of compressed renal parenchyma was identified. The renal capsule and the capsule of the locules were intact. The kidney from the third patient measured 11 × 5.5 × 4.7 cm. On sectioning, the cystically dilated renal pelvis had a smooth white or pink inner surface. Attached to the inner surface of the cyst, there was a well demarcated 2.4 × 2.1 × 1.2 cm nodule that projected into the lumen of the cystic renal pelvis (Fig. 3). The capsule of the kidney and the capsule of the cyst were intact. The calyces did not show abnormalities.

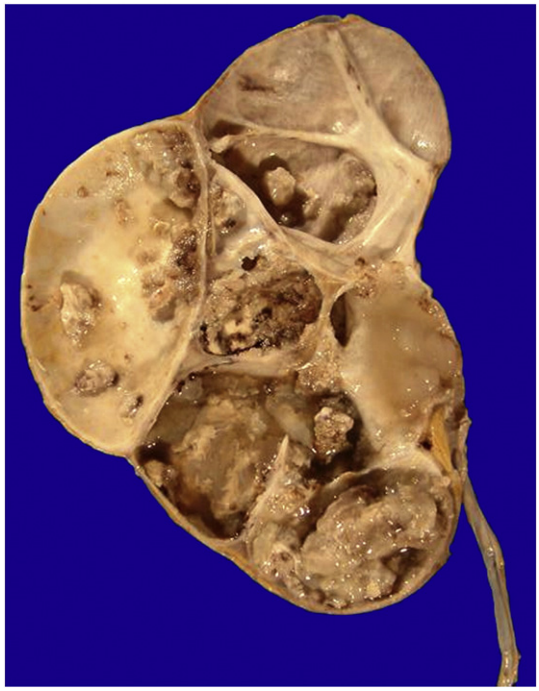

Fig. 1. A multiloculated cystic mass replaces the entire kidney.

2.3. Microscopic features

Multiple sections of the first 2 multiloculated cystic renal neoplasms showed similar histologic features. The locules were lined by a single layer of columnar cells with pseudostratified ovoid or elongated hyperchromatic basally placed nuclei. Few goblet cells were mixed with the columnar cells. In some areas, small papillary structures were identified. The nodules attached to the inner surface consisted of tubular (70%) and villous structures (30%) lined by columnar cells with intestinal phenotype (tubulovillous adenoma) (Figs. 4 and 5). In the lumen of the locules, there was abundant extracellular mucin, which contained fragments of columnar epithelium with low-grade dysplasia (Fig. 6). The nodule of the third case showed predominant

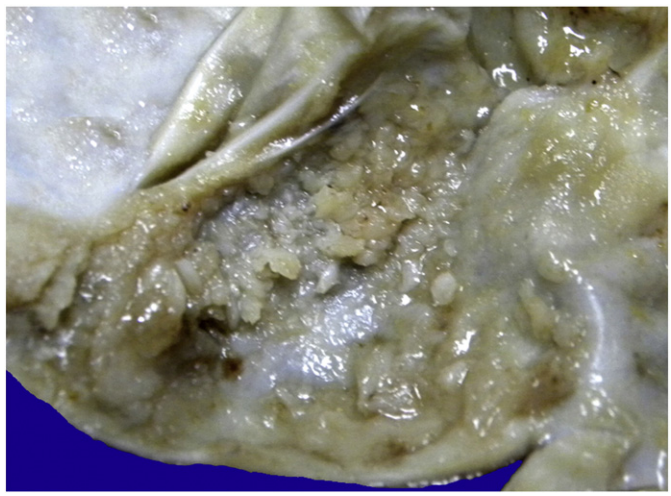

Fig. 2. The inner surface of a multiloculated mucinous cystadenoma of the kidney contains granular material and small nodules.

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