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Case Report

Parachordoma/myoepithelioma of the kidney: first report of a myxoid mimicry in an unusual location

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Abstract: We report a case of parachordoma (or myoepithelioma) of the right upper kidney in a 56 year-old male patient. Light microscopic features of the tumor exhibited epithelioid, glomoid, and spindle cells with eosinophilic and vacuolated cytoplasm as well as round to oval nuclei. These cells were embedded in a myxoid and hyaline stroma separated by a fibrous tissue with minimal cellular atypia and a few small nucleoli. Immunohistochemically, the tumor cells were immunoreactive for epithelial membrane antigen, calponin, vimentin, S-100, and type-IV collagen. All kidney and adrenal were resected, and the patient was carefully followed up. During the 11 months follow-up, recurrence and metastases were not observed. To our knowledge, this study is the first to document a case of parachordoma/myoepithelioma of the kidney. We add this new case to existing tumors and discuss its distinction from other types.

Keywords: Kidney, myoepithelioma, mixed tumor, parachordoma, myxoid

Introduction

Myoepithelioma/parachordoma/mixed tumor is a rare peripheral soft tissue tumor. Laskowski first presented parachordoma, and Dabska named and described this tumor in detail in 1977 [1]. Parachordoma is considered a unique entity by some pathologists [2], whereas others reported that this tumor is probably from soft tissue myoepithelioma/mixed tumor [3]. WHO fascicle listed this tumor as myoepithelioma/parachordoma/mixed tumor with uncertain intermediate soft tissue tumor before 2013 [4]. In 2013, WHO fascicle listed this tumor in the tumor of uncertain differentiations, renamed its myoepithelioma/myoepithelial carcinoma/mixed tumors and confirmed the biological behaviors. Parachordoma is also defined as myoepithelioma/myoepithelial carcinoma/mixed tumors [5]. These tumors show a reticular or trabecular growth pattern with myxoid, cartilaginous or hyalinized stroma. Tumor cells range from epithelioid to spindled and contain uniform nuclei with eosinophilic to clear cytoplasm. Myoepithelial carcinomas show similar histological features, in addition to the presence of nuclear atypia and together with a high mitotic rate, tumor necrosis.

This study first reports a case of parachordoma/myoepithelioma arising from the renal and describes its histopathological and immunohistochemical features. We also briefly addressed the differential diagnosis raised by this tumor in this uncommon location and with literature review.

Materials and methods

The case was obtained from the hospital consultation files of the Department of Pathology, Shihezi University School of Medicine, XinJiang China. This study was approved by the institutional ethics committee at the First Affiliated Hospital of Shihezi University School of Medicine and conducted in accordance with the ethical guidelines of the Declaration of Helsinki. Clinical information and radiological details were obtained from case files and electronic medical record files and electronic medical records. Two senior pathologists reviewed

Table 1. Immunohistochemical reagents and source

Antibodies	Clone	Vendor	dilution
CK	AE1/AE3	DAKO	1:100
CK7	OV-TL	DAKO	1:200
CK19	RCK108	DAKO	1:100
CK20	Ks20.8	DAKO	1:100
CK8/18	CAM5.2	DAKO	1:200
CK1/10	34βE12	DAKO	1:100
EMA	E29	DAKO	1:200
Vimentin	V9	DAKO	1:300
S-100	Polyclonal	DAKO	1:500
IV Collagen	C1V22	DAKO	1:100
CD34	QBend10	DAKO	1:200
CD10	56C6	DAKO	1:100
CD117	Polyclonal	DAKO	1:500
Calponin	CALP	DAKO	1:300
SMA	1A4	DAKO	1:200
desmin	DER111	DAKO	1:200
P63	4A4	DAKO	1:200
GFAP	GFAP	DAKO	1:200
Melanoma	HMB45	DAKO	1:200
Inhibin	R1	DAKO	1:300
D2-40	D2-40	DAKO	1:200
TFE3	Polyclonal	Santa Cruz	1:400

CK, cytokeratin; EMA, epithelial membrane antigen; SMA, smooth muscle actin; GFAP, Glial fibrillary acidic protein.

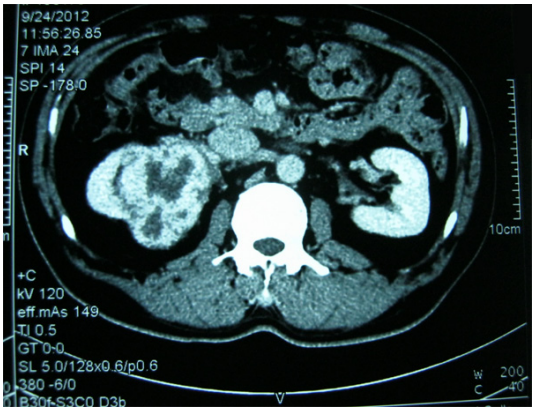

Figure 1. Computed tomographic scan of the abdomen. A mass is well-circumscribed in the upper kidney.

all original slides, including hematoxylin-eosin (HE) and immunohistochemistry staining, from each case. Immunohistochemical studies were performed on 4 μm-thick unstained sections generated from formalin-fixed, paraffin-embedded tissue. All of the immunohistochemical reactions were carried out in an automated immunostainer (LEICA Bond-Max autostainer, Leica; Germany). Antibody markers include cytokeratin AE1/AE3, cytokeratin CAM5.2, epithelial membrane antigen (EMA), cytokeratin 7, cytokeratin 20, cytokeratin 19, vimentin, S-100, CD34, CD10, CD117, collagen IV, calponin, SMA, desmin, p63, GFAP, TFE3, melanoma, D2-40, and α-inhibin. **Table 1** lists the details of the markers identified by the various antibodies. Appropriate positive and negative controls were also included.

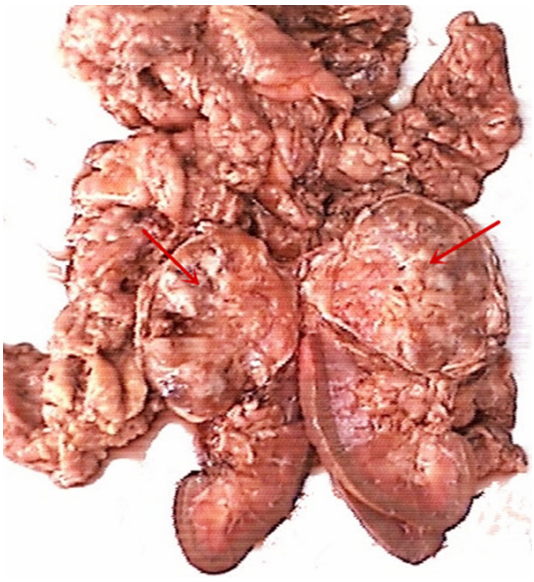

Figure 2. Cut surface of the tumor. The tumor was lobulated and solid with a grayish-white color.

Results

Case report

A 56 year-old man complained of pain and swollen right abdominal and iliopsoas muscle of about one month, and the detailed location was uncertain. He first noticed the same symptom over approximately 8 years, and underwent B-ultrasonic examination at a hospital in Shihezi, Xinjiang in 2004. A mass in the right upper kidney was observed measuring approximately 4 cm × 2 cm, but the patient did not undergo treatment. In 2012, a neoplasm of 7 cm in diameter was observed in the right upper kidney by CT (**Figure 1**), which we diagnosed as parachordoma (or myoepithelioma). These lesions were typically managed by resection of right renal and adrenal, and long-term

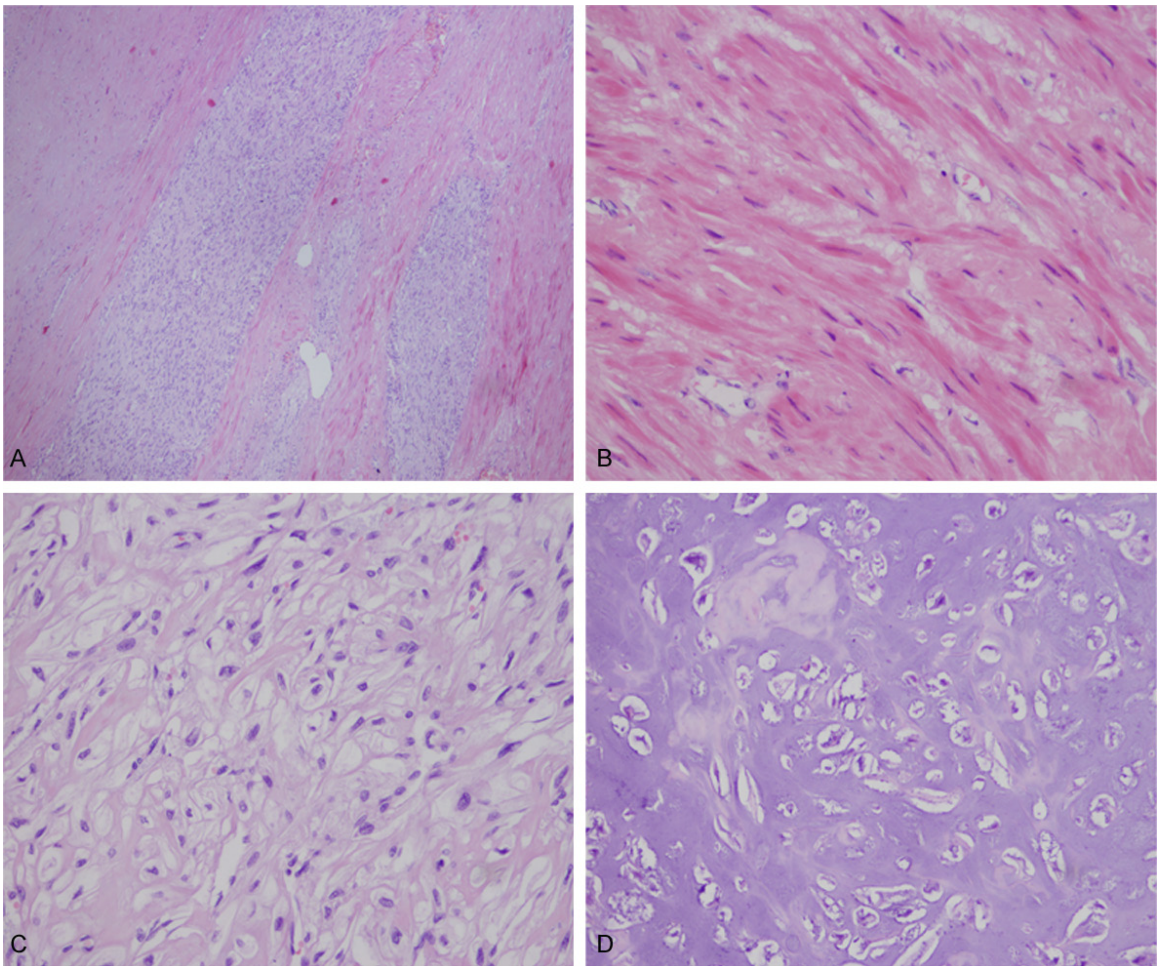

Figure 3. A: Tumor showing multinodular masses separated by broad collagen bands (HE 20×). B: Tumor showing spindle cells (HE 200×). C: Tumor arising from epithelioid cells; some were vacuolated (HE 200×). D: Tumor showing a myxochondroid stroma (HE 200×).

follow up studies were also carried out. At the time the present case report was written, the patient was alive with no signs of tumor recurrence and metastasis.

Pathological findings

The surgical specimen grossly consisted of a kidney and an adrenal covered by a thick fat. On a cut surface (**Figure 2**), the tumor was located on the upper renal with a size of 7 cm × 6 cm × 6 cm and well-demarcated from the surrounding tissues. It was firm and lobulated, and some regions contained translucent, cartilaginous-like tissue with a grayish-white color. Neither necrosis nor hemorrhage was observed in the tumor.

It microscopically forms a circumscribed and multinodular growth tumor (**Figure 3A**). The tumor was composed of round and spindle cells with eosinophilic and vacuolated cytoplasm (**Figure 3B**, **3C**). These cells were arranged in clusters, chains, nodules, and whorl formations, but did not exhibit a glandular architecture. These cells were embedded in a chondromyxoid and hyaline stroma (**Figure 3D**), separated by a fibrous tissue. Bland round to oval nuclei were found, and mitotic figures were rare. Necrosis or vascular invasion was absent.

Immunohistochemical findings

The present case immunohistochemistry revealed the expression of EMA (**Figure 4A**), CK8/18 (**Figure 4B**), S-100 (**Figure 4C**), calponin (**Figure 4D**), vimentin (**Figure 4E**), type-IV collagen, and CD117 by tumor cells with negative staining for CK1/10, CK20, CK7, CK19, SMA, desmin,

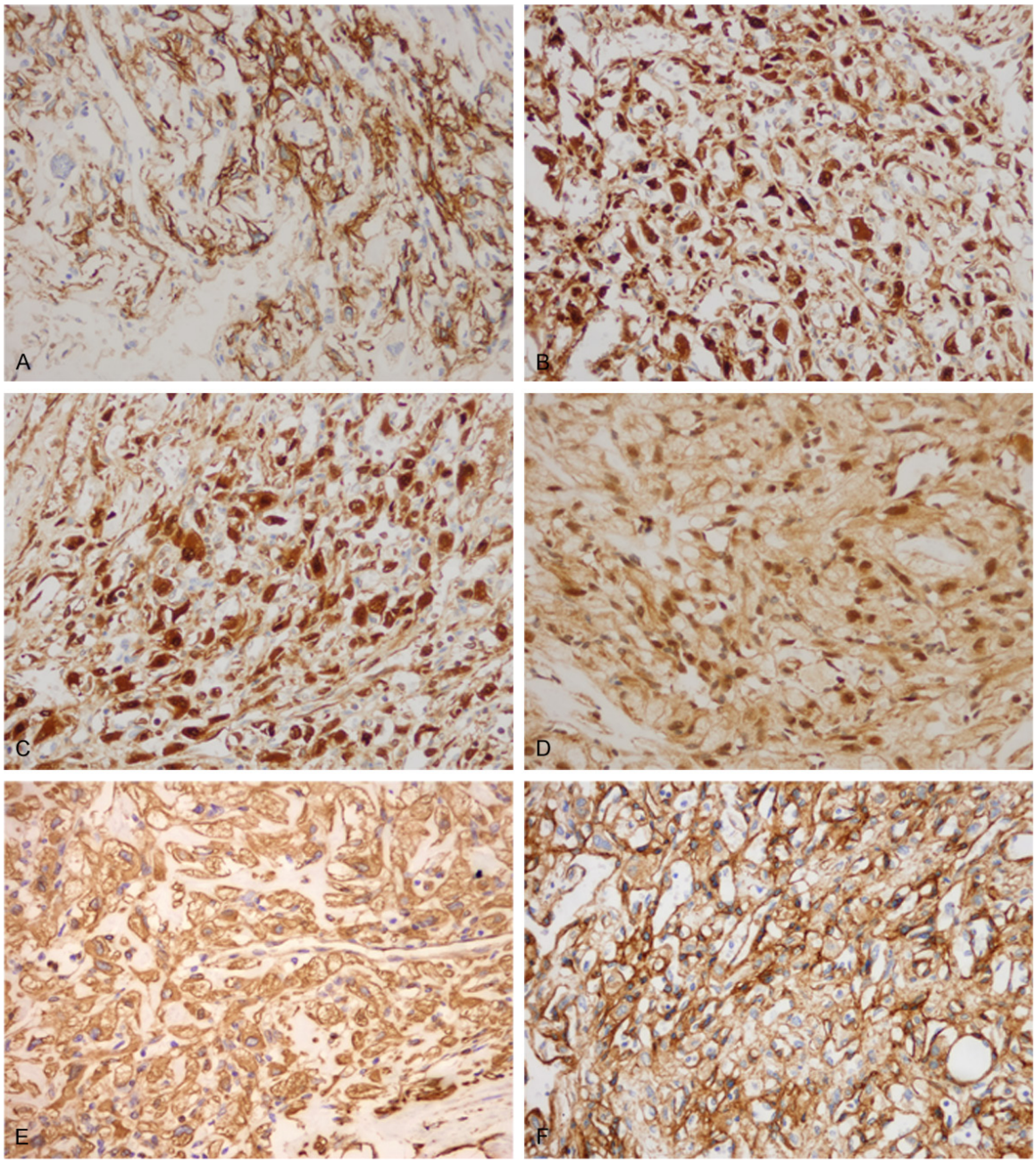

Figure 4. Immunohistochemistry exhibited positive expression of the present renal tumor: A: EMA (200×), B: CK8/18 (200×), C: S-100 (200×), D: Calponin (200×), E: Vimentin (200×), F: Collagen IV (200×).

CD34, CD31, CD10, GFAP, P63, TFE3, melanoma, D2-40, and α-inhibin. Type-IV collagen embraced groups of tumor cells in a nest-like appearance (**Figure 4F**).

Discussion

A renal tumor with rare morphological and immunohistochemical features was reported. Microscopic examination revealed a circumscribed tumor with clusters, unusual whorls, nodules, and chain formation of round, spindle, and vacuolated cells in a myxoid stroma and separated by a fibrous tissue. Differential diagnosis for these tumor histologic findings include renal cell carcinoma, other renal tumors, chondroid lipoma, myxoid liposarcoma, extraskeletal myxoid chondrosarcoma, chordoma, salivary gland pleomorphic adenoma.

Renal cell carcinoma in the adult clear cell renal cell carcinoma (CCRCC) is the most common histological variant. CCRCC section exhibited a typically golden color because its cells are rich in lipid content. It usually had necrosis, bursal lumen, and hemorrhage. Nests of clear cells were separated by vascular network. CCRCC immunohistochemical findings showed expression of CK and CD10. Papillary renal cell carcinoma and mucinous tubular and spindle cell renal cell carcinoma had a prominent tubular architecture [6]. Chromophobe cell carcinoma showed a mixture of clear and eosinophilic-type cells, which were almost associated with Xp11.2 translocations/TFE3 gene fusion renal cell carcinoma (Xp11.2RCC) positive expression TFE3 [7]. Collecting duct carcinoma was characterized by an invasive border, a tubulopapillary architecture, and high-grade cytologic features. Medullary renal cell carcinoma exhibited a tubular form with infiltration by neutrophils and had a rhabdoid-like morphology [8]. Other renal tumors, both benign and malignant, considered in the differential diagnosis are described in this study. Based on morphology, an adult renal myoepithelial hamartoma was excluded [9]. Metanephric adenoma is composed of low cuboidal-shaped tumor cells, and cytoplasm and tubular arrangement is rare. In addition, myxoid stroma is lacking, and the epithelial cells show a basophilic cytoplasm. The lesion component of spindle cells may be a sarcomatoid renal cell carcinoma, but mitotic figures are rare, such as benign or low-grade tumor. For renal cell carcinoma, other renal tumors can be excluded clinically, microscopically, and immunohistochemically.

Chondroid lipoma usually has a distinctive component, including lipoblasts. This tumor was often positive for S-100 protein, but usually negative for CK. Myxoid liposarcoma was easily excluded on the morphological basis of Sudan III stain, and the detected Fus-CHOP fusion gene break-apart rearrangement was positive. Vimentin and S-100 showed extraskeletal myxoid chondrosarcoma immunohistochemical expression, but all CK were negative. In addition, the balanced chromosomal translocation t (9; 22) (q22; q12), with breakpoint involving the EWS gene on chromosome 22q12 and the CHN gene on 9q22, is a characteristic for diagnosis of extraskeletal myxoid chondrosarcoma [10]. Parachordoma/Myoepithelioma has a wider variety of patterns than chordoma. Both tumors show similar immunohistochemical profile with epithelial marker and S-100 protein positively, but parachordoma/myoepithelioma negative for CK7 and CK19 is expressed in chordoma [3].

Salivary gland pleomorphic adenoma shows a degree of morphological diversity. The components are epithelial, myoepithelial, and mesenchymal cells or stromal element. The epithelial components include a variety of cell types including squamous, spindle, and clear cells. The mesenchymal-like cells are composed of myxoid, cartilaginous, or hyaline. Immunohistochemical study showed expression of CK, vimentin, CD10, S-100, and calponin. For similar morphological and immunohistochemical features, we carefully examined the clinical information and radiological details as well as the electronic medical record files and electronic medical records on salivary glands to exclude the possible primary pleomorphic adenoma/carcinoma from renal. Pleomorphic adenoma lymph node metastasis case is reported in literature [11]. A case was reported as a peculiar benign mixed renal neoplasm, which was discovered 1 year later as a salivary gland carcinoma from kidney [12]. The patient was followed up, and his ultrasound examination revealed an absence of salivary gland lesion.

If the aforementioned tumors are excluded, shared morphological and immunohistochemical features exist. Thus, the tumor was phenotypically consistent with parachordoma/myoepithelioma. We reviewed approximately 17 cases from the English literature published from 2007 to 2013 (**Table 2**) and 45 cases of parachordomas from 1977 to 2007 [13]. Parachordoma incidence ranged in age from 4 years to 86 years, which usually occurred in adults (57/62, 91.9%). A significant number of cases increased in children <10 years old, with five cases (5/62, 0.81%). They show equal distribution between the sexes. Among the case reports that mentioned location (upper extremities > lower extremities > trunk > buttock), extremity was the most common location, with 36 of 62 (58%) documented cases in the extremities, 8 of 62 (12.9%) cases in the trunk, and 4 of 62 (6.4%) cases in the buttock. One case occurred in the stomach (1/62, 1%), but no case in the kidney. Most tumors were benign. Of the 62 patients, nine (14.5%) and eight (12.9%) cases were reported to be recurrent

Table 2. Clinical and immunohistochemical reveals of parachordoma case reports

Cases (Year)	Age/Gender	Site	Recurrence	Metastasis/Fatal	Immunohistochemistry
[17] (2007)	65/F	thigh	NO	NO/NO	CK8/18+, EMA+, VIM+, S100+, GFAP+, calponin+, SMA+, CK1/10-
[18] (2007)	65/F	Gastric serosa	NO	NO/NO	CK+, EMA+, S100+, VIM+, BU-, CD117-, CD10-, GFAP-, calponin-, calretinin-, P63-
[13] (2008)	60/F	Arm	NO	NO/NO	CAM5.2+, S100+, CK19-, CK-, EMA-, MSA-, desmin-, CEA-
[19] (2009)	6/F	Forearm	NO	Metastasis/NO	CK+, VIM+, S100+, GFAP+, CgA+, SMA+, HMB45-, Syn-, Ki-67 (10%)
[20] (2009)	NK	Iliopsoas	NO/	Metastasis/NO	Unknown
[21] (2009)	76/M	Hand	2 years	NO/NO	Unknown
[22] (2010)	63/M	Skull	NO	NO/NO	Unknown
[23] (2010)	17/M	Buttock	Yes	NO/NO	CK+, CAM5.2+, VIM+, S100+, EMA+, CollIV+, CD34, CD31-, CD99-, FactorVIII-, D2-40, SMA-, desmin-, HMB45-, MelanA-
[24] (2011)	31/M	Wrist	One month	NO/NO	CK+, VIM+, S100+, EMA+, CK8/18+, CollIV+, desmin-
[25] (2011)	67/F	Arm	NO	Metastasis/No	CK+, S100+
[26] (2011)	28/M	Shoulder	One year	Unknown/NO	VIM+, S100+, EMA+, CK+, CK8/18+, GFAP-, HMB45-, CgA-, MelanA-
[14] (2011)	48/F	Presacral	NO	NO/Renal failure	CK+, VIM+, S100+
[27] (2012)	48/F	Pelvic	NO	NO/NO	VIM+, S100+, CollIV+, CD99+, EMA-, desmin-, actin-, CK5/6-, calretinin-, WT-, Ber-EP4-, Alpha-inhibin-, CD34-
[4] (2012)	44/M	Index finger	NO	NO/NO	CK+, VIM+, S100+, EMA+, CAM5.2+, CD34-, GFAP-, Ki-67 (<15%)
[28] (2012)	46/F	Peripheric um	NO	NO/NO	CK+, VIM+, S100+, EMA+, desmin-, SMA-, HMB45-, MelanA-, CD117-
[29] (2013)	32/M	Arm	NO	NO/NO	CAM5.2+, S-100+, CK+, EMA+, CD31-, CD34-, actin-, desmin-, MYOD1-, HMB-45-
[30] (2013)	42/M	Chest	NO	NO/NO	VIM+, S-100+, +, EMA+, CK+, CD31 (-)

F, women; Male, man; VIM, vimentin; CollIV, Type-IV Collagen; Syn, synaptophysin; BU, brachyury.

and metastatic, respectively. The results also showed that six of 62 (9.6%) cases were fatal, but one presacral parachordoma caused intestinal obstruction in a patient with renal failure [14]. However, extrapolating a recurrence rate, metastases, and fatal rate from these numbers is inaccurate because follow-up time in many patients in the case reports was insufficient. Therefore, patient follow-up and wide mass resection were essential to exclude recurrence.

A review of studies on the absence of reported parachordoma in the kidney has not been reported. However, the latest WHO classification of soft-tissue tumors indicated that parachordoma has the same description as soft tissue myoepithelioma/myoepithelial carcinoma/mixed tumors. The largest published series consisted of 101 cases myoepithelial tumors [15], with tumors found in soft tissue, its occurrence in a kidney site has not been reported.

Immunohistochemistry (**Table 2**) showed that all cases were positive for vimentin (10/10, 100%), S-100 (14/14, 100%), type-IV collagen (3/3, 100%), CK8/18, and CAM 5.2 (7/7, 100%). Almost all cases were positive for CK (11/12, 91%) and EMA (9/10, 90%). Calponin (1/2, 50%) and GFAP (2/4, 50%) were positive in half of the cases. A subset of tumors was positive for CD99 and SMA. However, CK19, desmin, CD10, CD34, brachyury, D2-40, HMB45, melan-A, and α-inhibin were consistently negative.

Among the possible different diagnoses excluded, reviewed, and compared, the morphological and immunohistochemical features showed that the final diagnosis of this case was parachordoma (or myoepithelioma). A similar case description existed [16], myxoid renal tumor with myoepithelial differentiation mimicked a salivary gland pleomorphic adenoma. The author did not diagnosis only concluded that a definite classification and histogenetic interpretation of this previously unreported tumor type awaits description and genetic analysis of similar cases.

In conclusion, we first diagnosis a case of parachordoma (or myoepithelioma) of the kidney and briefly addressed the differential diagnosis raised by this tumor in this uncommon location and literature review. Once a large series of these tumors are reported and characterized by genetic studies, we may have a better understanding of these tumors in the kidney.

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Disclosure of conflict of interest

None.

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