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Case Report
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Glomus Tumor in Kidney: A Case Report and Review of Literature

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Abstract

Background: Glomus tumor are uncommon tumors of mesenchyme consist of cells of the normal glomus body, smooth muscle and vasculature. Glomus tumor often occurs in the skin of distal extremities and visceral organ invasion is rare. We present a 54-year-old female with an accidentally found renal tumor.

Case Report: A 54-year-old female with past history of dyslipidemia and hypothyroidism. Asymptomatic renal tumor was noted under computed tomography accidentally. The image showed a heterogeneous tumor around 4 cm in diameter with calcification and septa in the upper pole of the left kidney. Due to the suspicion of malignancy, she received surgical resection of renal tumor. Immunohistochemical stains were performed with resected tumor cells, which revealed immunoreactivity for Smooth Muscle Actin (SMA) and Muscle Actin. Pathology report showed renal glomus tumor. No adjuvant therapy was given to this patient. There was no recurrence during follow-up so far.

Conclusion: Renal glomus tumor is rare and there are few cases reported before and most cases are benign tumor. Malignant glomus tumor is relatively scarce and the diagnosis is based on pathological characteristics. Mainly treatment of renal glomus tumor is surgical resection, either partial or radical nephrectomy. This case demonstrated a benign renal glomus tumor under pathological examination after partial nephrectomy. Prognosis of renal glomus tumor is ideal. The patient was disease-free as yet without recurrence.

Keywords: Glomus tumor; Urinary tract; Kidney; Renal

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Abbreviations

C/T: Chemotherapy; CT: Computed Tomography; H&E: Hematoxylin and Eosin; HPF: High-Power Field; MSA: Muscle Specific Actin; RT: Radiotherapy; SMA: Smooth Muscle Actin

Introduction

The glomus tumor, which resembles glomus body, which is an uncommon, perivascular neoplasm. Glomus tumors are rare soft tissue tumors that have similar incidence in both genders. Mostly, diagnosis is made in people between 20 to 40 years old and symptoms may precede years before [1]. There are several variants of glomus tumor besides classic histology, including glomangioma, glomangiomyoma and glomangiomatosis. Glomus tumor often occurs in the skin of distal extremities like subungual region of finger, palm, wrist, forearm and foot [1]. Occurrence in the visceral organ is rare, including gastrointestinal tract, lung, mediastinum, pancreas and kidney [2]. There is no definite guideline on treatment of renal glomus tumor.

In this report, we describe a 54-year-old woman who presented with a rare primary glomus tumor in the kidney, along with a literature review.

Case Presentation

A 54-year-old female with past history of dyslipidemia and hypothyroidism.

A left renal mass around 4 cm in diameter with calcifications and septa in the upper pole was noted under Computed Tomography (CT) accidentally (Figure 1). The tumor was partially enhanced, heterogenous and border was clear. She denied hematuria nor palpable mass except occasionally left flank pain during these days.

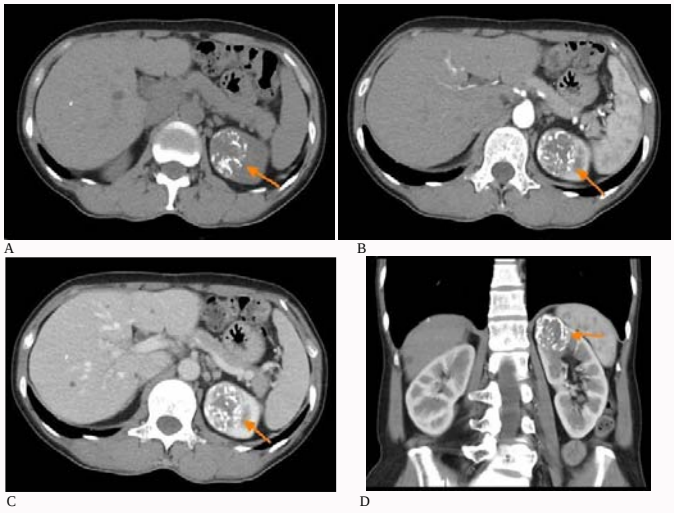

Figure 1: CT image of our cases: (A) Pre-contrast CT showed heterogenous renal tumor with calcification over upper pole (B) Arterial phase (C) Venous phase (D) Coronal view.

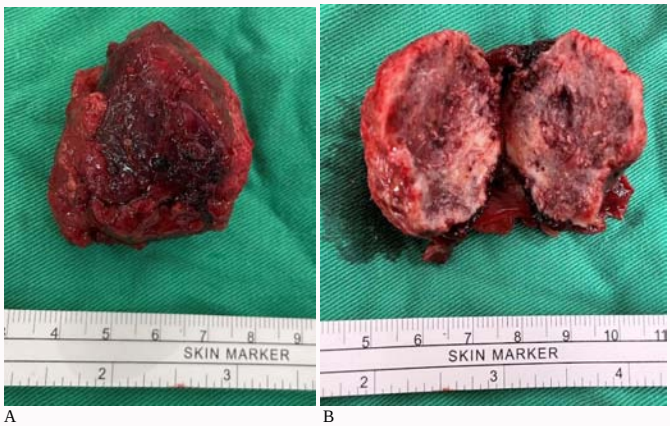

Figure 2: Tumor specimen: (A) Gross tumor specimen (B) Cross section of the tumor.

After fully discussion with the patient, she decided to receive laparoscopic partial nephrectomy under the suspect of renal cell carcinoma. The operation course was smooth and no acute complication was noted. Macroscopically, the resected tumor was measured 45 mm × 30 mm × 30 mm in size and was confined to the renal parenchyma.

From histopathological examination, immunohistochemical stains of tumor showed immunoreactivity for Smooth Muscle Actin (SMA) and Muscle Actin (Figure 2). No expression within the tumor cells for Desmin, CK, PAX-8, INSM-1, Synaptophysin, CD31, CD34, HMB-45 nor GLUT-1. Glomus tumor was impressed based on immunohistochemical stains features mentioned above. Other part excised along with the tumor showed chronic pyelonephritis microscopically. Discharge was arranged for the patient on the sixth day after the operation.

As of now, no renal function impairment was noted during follow-up after the operation. Computed tomography followed four months after the operation showed no recurrence of tumor. The patient received follow-up with CT annually and was free of disease at sixteen months after the operation.

Discussion

The normal glomus body is a specialized form of arteriovenous anastomosis that regulates heat. The most distribution site of glomus body is extremities, including digits and palms and it is usually located in the stratum reticularis of the dermis. Based on report from Masson, the normal glomus body is composed of afferent arteriole and turns into four preglomic arterioles, which blend with an irregular, thick-walled segment known as the Sucquet-Hoyer canal containing the arteriovenous anastomosis [3].

Glomus tumors are tumors of mesenchyme that resemble the normal glomus body with smooth muscle and vasculature. Masson firstly proposed the concept of glomus tumor in 1924, according to three cases diagnosed as upper extremity tumor with intermittent sharp pain [3]. Symptoms subsided after tumor excision. He found out the tumor structure was similar to glomus body and suggested that it was related to hyperplasia or overgrowth of glomus body.

According to report from Mayo Clinic, estimated incidence of glomus tumor was 1.6% among 500 consecutive soft tissue tumors. Both genders have equivalent incidence, while patient with subungual lesions are mostly female. Most glomus tumors are diagnosed on

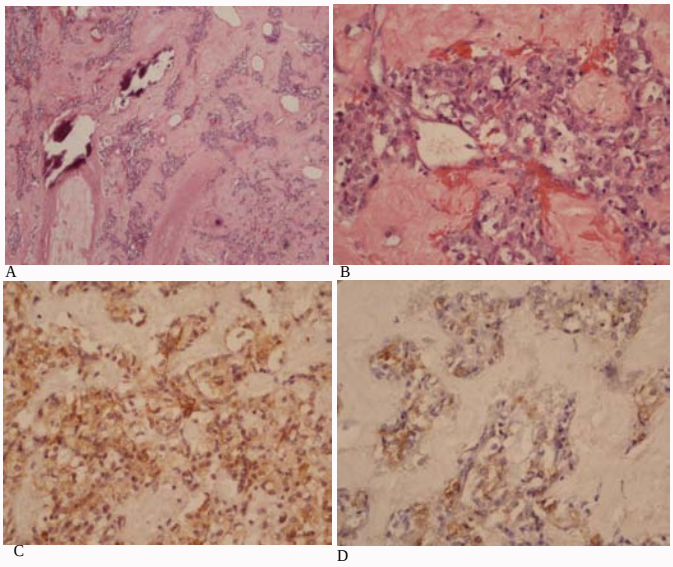

Figure 3: Immunochemical stain of tumor specimen (A) Hematoxylin and Eosin (H&E) stain 40x (B) H&E) stain 200x (C) Immunoactivity for muscle actin (D) Immunoactivity for smooth muscle actin.

Table 1: Review of previous reported renal glomus tumor.

Reference	Age, Sex	Size (cm)	Location of kidney	Image findings	Treatment	Positive immunochemical stain	Follow-up
Benign Glomus Tumor							
Siddiqui et al. [7]	55, Female	2	Left lower pole	Not stated	Partial	SMA, vimentin	Not stated
Heraw et al. [6]	53, Female	2.5	Right ureteropelvic junction	CT: Solid mass with hydronephrosis Nonfunctioning kidney and atrophy of the surrounding parenchyma	Radical	SMA, collagen type 4	Free of disease at 6 months
Al-Ahmadie et al. [8]	36, Male	3.3	Right anterior interpole	Sonography: Peripherally echogenic and centrally hypoechoic, confined to the renal capsule	Partial	SMA, muscle actin	Free of disease at 62 months
Al-Ahmadie et al. 2007	81, Male	4	Right lower pole	Not stated	Radical	SMA	Free of disease at 24 months
Al-Ahmadie et al. 2007	48, Male	7.3	Right mid/ lower pole	CT: Moderately enhancing, multilobulated	Radical	SMA	Free of disease at 33 months
Sasaki et al. [2]	62, Male	1.8	Left lower pole	CT: Enhanced lesion	Partial	SMA, vimentin, CD57, collagen type 4	Free of disease at 2 months
Gravet et al. [11]	60, Male	2.5	Left upper pole	CT: Enhanced, exophytic lesion	Partial	SMA, vimentin	Free of disease at 8 months
Present report	54, Female	4	Left upper pole	CT: Calcifications and septa	Partial	SMA, muscle actin	Free of disease at 16 months
Atypical and Malignant Glomus Tumor							
Gill and Van Vliet, [9]	46, Male	8.7	Right lower pole	CT: Exophytic, irregular peripheral enhancement, with septations, and central necrosis	Radical	SMA, MSA, CD34, bcl-2, vimentin, synaptophysin	Free of disease at 15 months
Lamba et al. [10]	44, Male	Metastasis primary tumor size unknown	Posterior right kidney Metastasis to spine and pelvic bones	CT: Mixed cystic and solid components arising from the posterior right kidney Multiple osseous metastases involving the spine and pelvic bones	Palliative RT + C/T	SMA, CD34, vimentin, collagen type 4	Died of disease at 6 months
Lai et al. and Chen et al. [12,13]	46, Male	3.7	Right upper pole	Sonography: Slightly hyperechoic CT: Contrast-enhanced, heterogeneous	Radical	Not stated	Free of disease at 6 months
Li et al. [4]	31, Female	16	Right	CT: Heterogeneous mass with an area of central necrosis	Radical	Vimentin, collagen type 4	Recurrence after 7 years of follow-up Died of disease at 13 years
Li et al, 2018	33, Female	9.7	Left with renal vein and IVC thrombus and tricuspid valve vegetation	Not stated	Radical	Vimentin, MSA	Not stated
Li et al, 2018	55, Male	1.5	Left	Sonography: cystic mass	Partial	SMA, collagen type 4	Not stated
Zhao et al. [5]	8, Female	5	Right upper pole	CT: Well-demarcated, hypodense, solid, with homogeneous contrast enhancement	Partial	SMA, MSA, vimentin, collagen type 4, CD34, renin	Free of disease at 16 months

people aged between 20 and 40 with symptoms always precede before the diagnosis.

Subungual region was the most seen site of glomus tumor, while other extremities region was also usual, including foot, forearm, palm and wrist. Few literatures may also have reported glomus tumor in gastrointestinal tract, the genital organ, oral cavity, trachea, mediastinum, heart and lymph nodes. So far, there are several cases of renal glomus tumor and were summarized in Table 1 [2,4-13]. Classically, glomus tumors are solitary, while Okada O et al. and Sawada S et al. ever reported multiple subungual glomus tumors in patients with Neurofibromatosis 1 (NF1) [14,15]. Symptoms of glomus tumor often disproportionate with size. Some reported radiating pain away from the lesion and may be induced by changes in temperature, especially to cold, and tactile stimulation of even minor degree.

Mostly, renal glomus tumors behaved as benign tumor, while malignant glomus tumors were also reported in some case reports [6,8,11]. There are no specific symptoms among these cases, include flank or abdominal discomfort and microscopic hematuria. Overall, most cases were diagnosed accidentally under image examination.

Radiology image is widely used for differential diagnosis of renal mass. Currently, due to low incidence rate and radiologic characteristics, it is not easy to diagnose glomus tumor of the kidney depends on only image [16]. Many previous reported cases showed enhanced, heterogenous lesion under CT with clear border. In our case, image revealed diffused calcification and malignancy was suspected. During the operation, firm texture of tumor was noted, which was different from common renal tumor.

Glomus tumor usually occurred in middle-aged people. Zhao et al. ever reported one case of a young girl with past history of tuberous sclerosis complex diagnosed as renal glomus tumor at the age of age [5]. The patient received partial nephrectomy and was free of disease at 16 months of follow-up.

Tissue biopsy or pathology from surgery is necessary for confirming the diagnosis of glomus tumor. Immunohistochemical diagnosis is made based on immunoreactivity for both common muscle and smooth muscle actin regardless of different location of tumor [17,18]. Renal glomus tumor is distinguishable from renal cell carcinoma because it is positive for muscle markers while negative for epithelial markers including cytokeratin and epithelial membrane antigen. Immunoactivity of desmin of tumor was variable, ranging from focal positivity to no expression [9]. It is reported that peripheral/subcutaneous glomus tumor expressed CD34 positivity more frequently than visceral glomus tumor [19]. S-100 protein, various cytokeratin's and other markers including CD31, HMB-45, CD117, CD20, CD45, C56 and CD57 showed negative result mostly [6,7,18,20] (Figure 3).

Previous study has defined the criteria for possible malignancy, which includes (1) tumor size larger than two centimeter and deep location in kidney; (2) display of atypical mitotic figures; (3) significant nuclear grade and mitotic activity (5 mitoses/50 HPF) [21]. In our cases, tumor size was larger than two centimeter and located deep in upper pole of left kidney, while pathology report showed no obvious increased mitosis or necrosis. Surgical resection remained mainstay treatment in previous reported cases [2,6-9,11,13]. Mostly, the prognosis was acceptable, either pathological report showed atypical and malignant or benign glomus tumor. Lamba et al. [19], however, firstly reported malignant renal glomus tumor with pelvic bones and spine metastasis. Palliative chemotherapy and radiation therapy was given while poor response was noted. The patient passed away within six months after diagnosis. To our knowledge, there was only one malignant renal glomus tumor ever reported in our living district according to the criteria. The patient received radical nephrectomy and adrenalectomy for the tumor and he was free of disease at six months of follow-up [12].

In conclusion, renal glomus tumor is an uncommon tumor that encompassed unspecific clinical symptoms. It is challenging to distinguish renal glomus tumor from other renal tumors including renal cell carcinoma by symptoms, laboratory and image data. Definite diagnosis was always made based on histopathological examination. There are still limited data and consensus for treatment of renal glomus tumor due to scarcity. Because of the acceptable outcome and recurrence, surgical resection of the tumor may be considered, either partial or radical nephrectomy. Regular follow-up may be needed after primary treatment due to ever-reported recurrent case even though low recurrence rate was noted [4].

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