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Solitary fibrous tumor of kidney: a case report with extensive review of the literature

Bülent Kurt (*), Önder Öngürü (*), Ayhan Özcan (*), Ali Fuat Çiçek (*), Şeref Başal (**), Fatih Örs (***), Yıldırım Karslıoğlu (*), Ömer Günhan (*)

SUMMARY

A 49-year-old woman admitted to our hospital with a 3-month history of right flank pain without gross hematuria. Computed tomography scan of the abdomen showed that a 6x6x5 cm, contrast-enhanced mass was localized on the right renal pelvis. Histologically, a tumor composed of small ovoid to spindle shaped nuclei with indistinct cytoplasm was seen. Neoplastic cells showed diffuse immunoreactivity with CD34. It was diagnosed as solitary fibrous tumor. Solitary fibrous tumor of kidney is a very rare spindle cell neoplasm. It can mimic renal malignancy radiographically but almost all of renal solitary fibrous tumors are benign lesions.

Key words: *CD34, kidney, solitary fibrous tumor*

ÖZET

Böbreğin soliter fibröz tümörü: bir olgu sunumu ve literatürün detaylı gözden geçirilmesi

Kırk dokuz yaşındaki kadın hasta, gros hematüri olmaksızın 3 aydır süren sağ yan ağrısı şikayeti ile hastanemize başvurdu. Bilgisayarlı tomografide batında, sağ renal pelvis yerleşimli, kontrast tutan 6x6x5 cm boyutlarında kitle görüldü. Histolojik kesitlerde, küçük, iğsi-oval çekirdekli, sitoplazmik sınırları net olarak seçilemeyen hücrelerden oluşan tümör görüldü. Neoplastik hücreler CD34 ile kuvvetli immünreaktivite gösterdi. Lezyona soliter fibröz tümör tanısı konuldu. Böbreğin soliter fibröz tümörü, nadir görülen iğsi hücreli bir neoplazmdır. Radyolojik olarak renal maligniteleri taklit edebilir, ancak hemen tüm renal soliter fibröz tümörler benign lezyonlardır.

Anahtar kelimeler: *CD34, böbrek, soliter fibröz tümör*

Introduction

Solitary fibrous tumor (SFT) is a rare spindle cell neoplasm. It frequently arises from the serosal surface especially in pleural cavity (1). Extrapleural SFTs are very rare, and to our knowledge, there are only 28 cases of SFTs located in kidney. Clinically, this neoplasm can mimic renal malignancies. They are mostly benign lesions and characteristically exhibit diffuse CD34 positivity. We herein report an additional case of SFT originating from renal sinus and filling the renal pelvis, and summarize the clinical and pathological features of 29 previously reported SFT cases of the kidney.

Case Report

A 49-year-old woman presented with a 3-month history of right flank pain, without gross hematuria. Physical examination was unremarkable. Computed tomography scan of the abdomen showed that a 6x6x5 cm, contrast-enhanced mass was localized on the right renal pelvis (Figure 1a). Radical nephrectomy was performed subsequently. Specimen consisted of right kidney covered by perinephric adipose tissue. Macroscopic examination showed a well-circumscribed unencapsulated 6x6x5 cm mass originating from renal sinus and growing into the renal pelvis. The tumor had a firm, gray to white cut surface. Macroscopically, no necrosis, cyst formation, hemorrhage or renal vascular invasions were noted (Figure 1b). Microscopically, tumor was composed of hypercellular nodular areas and among them less cellular areas containing dense collagenous bands (Figures 2a, 2b). Neoplastic cells had small ovoid to spindle shaped nuclei with indistinct cytoplasm. Tumor cells were interspersed by a stroma including thin walled vascular spaces. There were no mitotic figures, necrosis or atypia.

Immunohistochemically, tumor cells were diffusely positive for vimentin and CD34 (Figure 3). Bcl-2 was

* Department of Pathology, Gulhane Military Medical Faculty
** Department of Urology, Gulhane Military Medical Faculty
*** Department of Radiodiagnostic Radiology, Gulhane Military Medical Faculty

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Reprint request: Dr. Bülent Kurt, Department of Pathology, Gulhane Military Medical Faculty, Etlik-06018, Ankara, Turkey
E-mail: bkurt_md@yahoo.com

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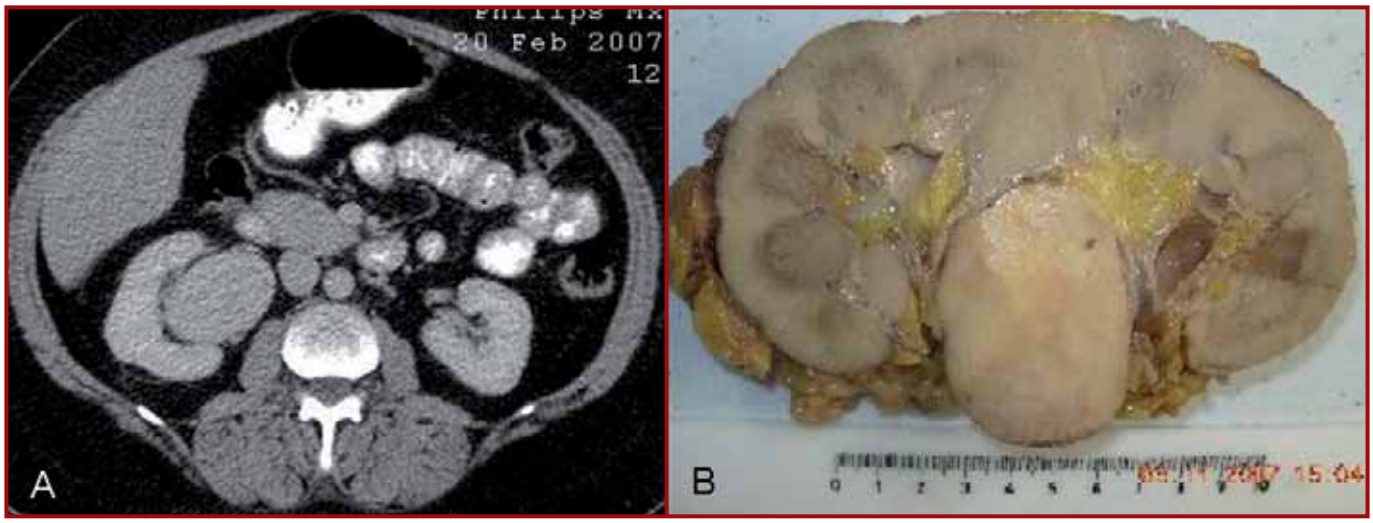

Figure 1. A. Enhanced mass localized on right renal pelvis, **B.** Well-circumscribed unencapsulated mass

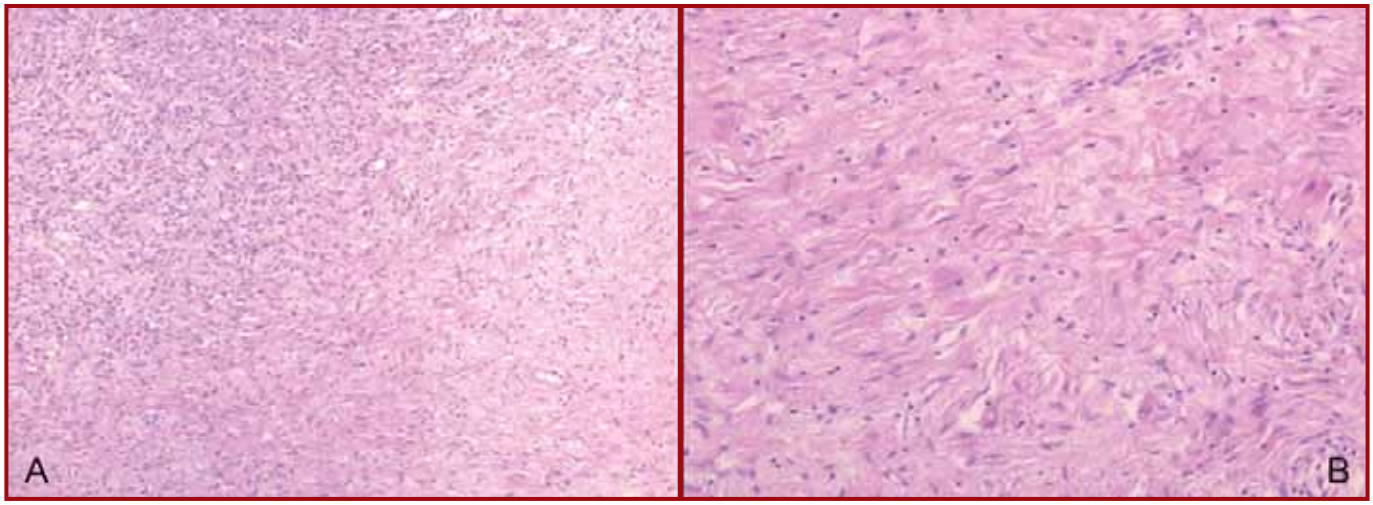

Figure 2. A. Hypercellular and hypocellular areas (HEx50), **B.** Less cellular dense collagenous bands containing areas (HEx100)

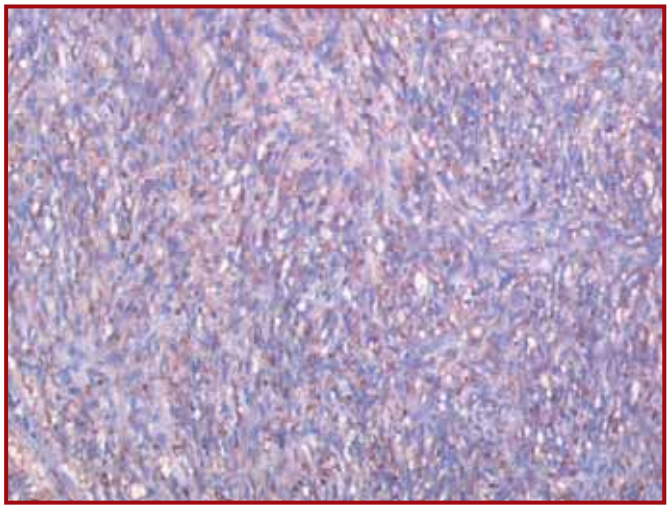

Figure 3. Strong CD34 positivity (x100)

weakly positive. Ki-67 (MIB-I) proliferation index was approximately 1%. Smooth muscle actin (SMA), desmin, S-100 protein, CD117 and CD99 were negative. It was diagnosed as SFT with the histological and immunohistochemical findings.

Discussion

Solitary fibrous tumor has first been described in 1931 (2). Etiology is unknown. Immunohistochemical and ultrastructural studies favor that the cell of origin for SFT is fibroblastic/primitive mesenchymal cells (3). They are most commonly described as pleural lesions, but many extrapleural locations have also been reported (1,4-7). A review about renal SFT has been recently published, which includes 19 cases. But, to our knowledge there are 29 cases in the literature (Table I) (8-31). First renal SFT has been reported by Gelb at al. in 1996 (8). Almost all renal SFTs are seen in adult life with a wide range (18-85) of age. Only one pediatric case has been reported in a 4-year-old boy (Case 26). Tumor predominantly affects female (F/M: 17/12). The most frequent clinical findings are abdominal

Table I. Clinical features of the previously reported solitary fibrous tumors of the kidney

No and References	Age/sex	Presentation	Location in kidney	Borders	Follow-up
1 Gelb et al. 1996 (8)	48/F	Flank mass and hematuria	Right, renal capsule	Well C.*	**
2 Fain et al. 1996 (11)	45/F	Upper abdominal pain	Right, intraparenchymal	Well C.	8 months
3 Fain et al. 1996 (11)	46/F	Abdominal pain	Right, intraparenchymal	Well C.	33 months
4 Fain et al. 1996 (11)	51/M	Chest pain	Left, intraparenchymal	Well C.	2 months
5 Hasegawa et al. 1996 (12)	64/M	Hematuria	Intraparenchymal	Well C.	8 months
6 Fukunaga et al. 1997 (13)	33/F	Lower abdominal pain	Right, renal peripelvis	Well C. No capsule	90 months
7 Fukunaga et al. 1997 (13)	36/F	Lower abdominal pain	Left, renal peripelvis	Well C. No capsule	1 year
8 Leroy et al. 2000 (15)	66/F	Abdominal pain, hematuria	Right, intraparenchymal	Renal invasion	9 months
9 Morimitsu et al. 2000 (14)	72/F	Not specified	U	Well C.	10 months
10 Wang et al. 2001 (16)	41/M	Hematuria	Left, intraparenchymal	Well C. encapsulated	48 months
11 Wang et al. 2001 (16)	72/M	Abdominal pain	Right, parenchymal	Well C No capsule	5 months
12 Yazaki et al. 2001 (17)	70/M	Not specified	Right, renal pelvis	Well C.	60 months
13 Kohl et al. 2006 (18)	85/F	Flank pain	Left, renal sinus	Well C.	U
14 Cortez-Gutierrez et al. 2001 (19)	28/F	Abdominal pain	Left, renal capsule	Well C.	12 months
15 Magro et al. 2002 (20)	31/F	Flank pain	Right, intraparenchymal	Well C.	8 month
16 Durand et al. 2003 (21)	35/M	U	Renal sinus	Well. C No capsule	U
17 Ibarguren et al. 2003 (22)	51/F	Abdominal mass	Bilateral	NE	NE
18 Bugel et al. 2003 (23)	60/F	NE	Right, intraparenchymal	NE	NE
19 Yamada et al. 2004 (24)	59/M	Not specified	Left, renal capsule	Well C. encapsulated	48 months
20 Gres et al. 2004 (25)	83/M	U	Right	U	U
21 Johnson et al. 2005 (26)	51/F	Back pain	Right, renal capsule	Well C.	U
22 Yamaguchi et al. 2005 (10)	51/F	Lower back pain	Left, renal capsule	Well C.	U
23 Koroku et al. 2006 (27)	18/F	Abdominal pain	Left, renal sinus	NE	15 months
24 Fine SM et al. 2006 (9)	76/M	Not specified	Left, intraparenchymal	Infiltrative	U
25 Alvarez et al. 2006 (28)	36/M	Flank colic pain	Right, renal capsule	NE	NE
26 Ferrari et al. 2006 (29)	4/M	Nonproductive cough	Right, intraparenchymal	Well C.	U
27 Bozkurt et al. 2007 (30)	51/F	Flank pain, hematuria	Left, parenchymal	Well C.	10 months
28 Znati et al. 2007 (31)	70/M	Back pain, hematuria	Left, parenchyma and perirenal tissue	Focal infiltrative	6 months
29 Current case	49/F	Flank pain	Right, renal sinus	Well C.	1 month

*: Well C: Well-circumscribed, **: Died after surgery, U: Unknown, NE: Not reported in the English literature

or flank pain. Occasionally abdominal/flank mass or hematuria can be seen. Some of tumors are detected incidentally. There is no predilection for left or right kidney. Only one case was bilateral (Case 17). Tumor was localized in various parts of kidney including parenchyma (13 cases), renal sinus (5 cases) and renal capsule (8 cases) (Table I).

Macroscopically, renal SFTs are classically well circumscribed, firm, solid and frequently lobular with gray to white cut surface. Myxoid, hemorrhagic and necrotic changes are occasionally observed. The tumor had a wide range of size (2-25 cm). Most of the cases were well circumscribed without cystic, hemorrhagic or necrotic changes. Only three cases had necrotic areas and two of them had infiltrating borders. One of these cases was diagnosed as malignant SFT (Case 24).

Microscopically, benign SFTs consist of hypocellular and hypercellular areas. Tumors are composed of spindle cells without an obvious growth pattern, so called “patternless pattern”. SFTs are tumors with variable cellularity and composed of a mixture of haphazard, storiform, short fascicular arrangements of bland spindle cells and collagenous bands. Spindle cells are not atypical and have little cytoplasm with indistinct borders. They are highly vascular tumors, and hemangiopericytoma like pattern can be seen in some areas. Myxoid changes and fibrosis may be ob-

served. Occasionally mast cells and lymphocytes can be seen. Mitosis is usually scarce (less than 3 per 10 high-power fields). Although most of SFTs are histologically benign, there are malignant SFTs reported in the literature (9). Malignant SFTs are highly cellular lesions. They show necrosis, focally or moderately nuclear atypia, numerous mitoses (more than 4 mitosis per 10 HPF) and/or infiltrative margins (10). We were able to find histological features for 22 of 29 renal SFTs in the literature (Table II). Almost all of the cases were benign with typical histopathological features for SFT. Only one renal SFT case had malignant features. Mitosis was rare (less than 3 mitosis per HPF) in benign renal SFTs. Malign SFT had high grade sarcomatous areas (more than 90% of tumor) composed of hyperchromatic and pleomorphic spindled cells surrounding staghorn like blood vessels with frequent mitosis and foci of tumor necrosis (9). This tumor also contained typical SFT areas. Focally aggregated mature fat cells were reported in one case of benign SFT (32).

Immunohistochemically, diffuse vimentin and CD34 positivity are seen in almost all of SFTs. CD34 and CD99 positivity are 90-95% and 70% in SFT, respectively (32). Furthermore, Flint and Van de Rijn have shown CD34 positivity in 77% and %78.5 of SFT cases, respectively in their studies (33,34). Bcl-2 expression in SFTs is found in various degrees (20-35%)

Table II. Pathologic features of the previously reported solitary fibrous tumors of the kidney

No and References	Size (cm)	Cyst	Hemorrhage	Necrosis	Mitosis	CD34	Vimentin	Bcl-2	CD99	SMA	Desmin
1 Gelb et al. 1996 (8)	3	-	-	-	Rare	+	U	U	U	-	-
2 Fain et al. 1996 (11)	6	-	-	-	Rare	U	+	U	U	-	-
3 Fain et al. 1996 (11)	7.2	-	-	-	Rare	+	+	U	U	-	-
4 Fain et al. 1996 (11)	4.5	-	-	-	Rare	+	+	U	U	-	-
5 Hasegawa et al. 1996 (12)	4.5	-	-	-	Rare	+	+	+	U	-	-
6 Fukunaga et al. 1997 (13)	3.5	-	-	-	Rare	+	+	U	U	-	-
7 Fukunaga et al. 1997 (13)	2	-	-	-	Rare	+	+	U	U	-	-
8 Leroy et al. 2000 (15)	9	-	-	-	Rare	+	+	U	Focal	-	-
9 Morimitsu et al. 2000 (14)	8	-	-	-	Absent	+	U	U	U	U	U
10 Wang et al. 2001 (16)	14	-	-	-	Rare	+	+	+	U	-	-
11 Wang et al. 2001 (16)	13	-	-	-	Rare	+	+	+	U	-	-
12 Yazaki et al. 2001 (17)	6	-	-	-	Absent	+	+	U	U	-	U
13 Kohl et al. 2006 (18)	3.5	-	-	-	Absent	+	+	+	U	-	-
14 Cortez-Gutierrez et al. 2001 (19)	15	-	-	-	Absent	+	+	U	U	-	-
15 Magro et al. 2002 (20)	8.6	-	-	-	Rare	+	+	Focal	Focal	-	-
16 Durand et al. 2003 (21)	17	NE	NE	NE	Rare	+	+	U	U	U	U
17 Ibarguren et al. 2003 (22)	L-25, R-2	NE	NE	NE	Absent	+	+	+	U	-	-
18 Bugel et al. 2003 (23)	11	NE	NE	NE	NE	+	+	NE	NE	NE	+
19 Yamada et al. 2004 (24	6.5	-	-	-	Absent	+	+	+	+	-	-
20 Gres et al. 2004 (25)	9	U	U	U	U	+	U	+	+	U	U
21 Johnson et al. 2005 (26)	11	-	-	-	Rare	Focal	+	+	Focal	-	-
22 Yamaguchi et al. 2005 (10)	10	-	-	-	Absent	+	+	+	+	-	U
23 Koroku et al. 2006 (27)	3	NE	NE	NE	NE	+	NE	NE	NE	NE	NE
24 Fine SM et al. 2006 (9)	12	-	-	+	Frequent	+	U	Focal	-	-	U
25 Alvarez et al. 2006 (28)	10	NE	NE	NE	NE	+	+	U	+	-	-
26 Ferrari et al. 2006 (29)	15	U	U	U	U	U	U	U	U	U	U
27 Bozkurt et al. 2007 (30)	3.7	-	-	-	Absent	+	+	+	+	-	-
28 Znati et al. 2007 (31)	15	+	-	+	Absent	+	+	+	+	-	U
29 Current case	6	-	-	-	Absent	+	+	Focal	-	-	-

U: Unknown, NE: Not reported in the English literature

in different studies (14,32). In a small group of SFTs (20-35%), SMA expression was also reported (32). S-100 protein, desmin and cytokeratin positivity were occasionally reported. There are no well-documented data for immunohistochemical staining pattern of renal SFTs. Almost all of renal SFTs (26/27) were diffusely positive for CD34. Only one case showed focal CD34 positivity (Case 21). Vimentin was diffusely positive in all cases (23/23). Nine cases exhibited CD99 positivity (Diffuse in 6 cases, focal in 3 cases). Two cases were negative for CD99. Bcl-2 was diffusely positive in most of the cases (11/14). Focal Bcl-2 expression was also shown in remaining cases (3/14). Neither SMA nor desmin expression was present in renal SFTs.

SFTs need to be distinguished from benign and malignant spindle cell tumors of the kidney. Differential diagnosis should include hemangiopericytoma (HPCs), fibroma, angiomyolipoma, leiomyoma/leiomyosarcoma, schwannoma, neurofibroma, perineurioma, monophasic synovial sarcoma and sarcomatoid renal cell carcinoma. Less than 30 primary renal HPC have been published in the literature. Most of them arise from the renal sinus and perirenal tissue. Distinction from hemangiopericytoma is sometimes difficult because the histological and immunohistochemical features of these two entities may show overlaps. Macroscopically hemorrhage is more common in HPC than in SFT. Although SFTs may consist hemangiopericytoma-like areas, presence of numerous, variably ectatic or compressed, thin-walled branching vessels having a staghorn configuration are more frequently seen in hemangiopericytoma (2). In HPC, neoplastic cells are usually diffusely dispersed and stromal hyalinization and varying cellularity are not usual in contrast to SFT (32). Immunohistochemically, most HPCs express CD34 but usually lesser degree than SFTs (2). So, there may be cases that can not be distinguished from each other. Our case showed diffuse and strong CD34 positivity (Figure 3). Renal medullary fibromas are usually smaller than 1 cm in diameter with parenchymal localization. They are negative for CD34. Lack of muscle and adipose tissue components distinguish SFT from angiomyolipoma. But sometimes SFTs may include adipose component. Leiomyomas/leiomyosarcomas show high percentage of SMA/desmin positivity and not express CD34. Schwannomas, neurofibromas and perineuriomas may occur in kidney. Schwannomas and neurofibromas express S-100 protein. Fifty percent of perineuriomas may show CD34 positivity but in contrast to SFT they exhibit also consistent EMA immunoreactivity. Sarcomatoid renal cell carcinomas show immunoreactivity with cytokeratins without CD34 expression. The monophasic synovial sarcoma may demonstrate some difficulties in the differential diagnosis. In contrast to SFTs, monophasic synovial sarcoma show at least focal immunoreactivity with cytokeratin and lack of CD34 expression.

Prognosis is generally good for SFTs. Most of them are non-recurring and non-metastazing tumors. Roughly 10-15% of SFTs are malignant (2). Only one case of previously reported 28 renal SFT has been reported as malignant (9), but unfortunately follow-up information was not available for this case. No recurrence or metastasis have been noted for the 19 cases with available follow-up (3-90 months).

In conclusion, we summarized clinical and pathological features of 29 cases of renal SFT. These tumors may mimic malignant tumors, clinically and radiographically. But most of renal SFTs are benign and their prognosis is very good. Only one case of malignant SFT of kidney has been reported. In differential diagnosis, the most useful tool is diffuse CD34 positivity to distinguish it from other spindle cell tumors of the kidney.

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