Clinical Case Report

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Deep benign fibrous histiocytoma occurring in the kidney

Case report

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Abstract

Rationale: Deep benign fibrous histiocytoma (BFH) is relatively rare in contrast to superficial BFH involving the skin. Moreover, it was extremely rare for deep BFH occurring in the solid organ. To our knowledge, so far, only one case of deep BFH of the kidney was reported in English literature.

Patient concerns: Herein, we report another case of deep BFH located in the kidney in a 88-year-old female. She was referred to our hospital for a severe pain in the right upper abdomen. Computed tomography revealed a round low-density shadow measuring 6 cm in the kidney.

Diagnosis: The lesion was diagnosed as a deep BFH of the kidney, as the tumor was histologically predominately composed of bland ovoid cells arranged in a storiform pattern.

Intervention: The patient underwent the total resection of the right kidney with the tumor in our hospital.

Outcomes: The postoperative course was uneventful. The patient was alive with no tumor recurrence or metastasis within 6 months of follow-up.

Lessons: We present another case of deep BFH of the kidney. Because of the rarity, the tumor may be poorly recognized. The typical storiform pattern in histology may be helpful for diagnosis. This report serves to remind that deep BFH is also a differential diagnosis for a tumor with storiform pattern in the kidney.

Abbreviations: BFH = benign fibrous histiocytoma, SFT = solitary fibrous tumor.

Keywords: deep benign fibrous histiocytoma, kidney, soft tissue tumor

1. Introduction

Benign fibrous histiocytoma (BFH) of the skin, also named as dermatofibroma, is the most common soft tissue tumor.[1–3] In contrast, deep BFH that occurred in subcutaneous and deep soft tissue is very rare.[4] It is extremely rare for deep BFH reportedly occurred in the retroperitoneum, mediastinum, oral cavity, and pelvis.[5–9]

Furthermore, deep BFH may show different histological features from the conventional BFH.[6,10] Conventional BFH is characterized by a variable number of spindle and/or rounded cells, arranged in peculiar storiform pattern admixed with inflammatory cells.[1–3] And a number of other uncommon variants including aneurysmal fibrous histiocytoma and epithelioid cell histiocytoma were reported.[1–3] Different from the ill-defined border of dermatofibroma, deep BFH usually presented as a well-circumscribed, but not encapsulated, mass.[4,6] Deep BFH frequently showed a positivity for CD34, leading to a confusion with other CD34-positive tumors.[6,10] Consequently, deep BFH may be poorly recognized and misdiagnosed by the pathologists.

To our knowledge, so far, only one case of deep BFH reportedly occurred in the parenchyma of kidney in English literature.[11] That is, Sakakibara et al[11] who described the first case of BFH that occurred in the left kidney of a 48-year-old man. Subsequently, Kobayashi et al[12] reported a case of deep BFH occurring in renal capsule, and not in the parenchyma. Then no other case was reported. Herein, we present the second case of BFH of the kidney. The tumor was located at the lower pole of the right kidney in a 88-year-old Chinese female.

2. Case presentation

2.1. Ethic approval

This study was approved by the Institutional Review Board of the Ethics Committee of China Medical University. Written informed

Editor: N/A.

This research was supported by National Natural Science Fund of China (81401885, funder: X-YL) and the funding from the Basic Scientific Project of University, Liao Ning Province, China (LQNK201705, funder: X-FL) to purchase antibodies and streptavidin-peroxidase system, respectively.

The authors have no conflicts of interest to disclose.

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Medicine (2019) 98:18(e15150)

Received: 29 September 2018 / Received in final form: 21 February 2019 / Accepted: 14 March 2019

http://dx.doi.org/10.1097/MD.0000000000015150

consent was obtained from the patient for publication of this case report and accompanying images and the study was performed in accordance with the Helsinki II declaration.

2.2. Clinical history

A 88-year-old female was referred to our hospital for a severe pain in the right upper abdomen, diverging to the right side of the waist and back. The patient then underwent an abdominal computed tomography scan. Computed tomography revealed that there were multiple nodular high-density shadows in the gall bladder and a round low-density shadow measuring 6 cm in the kidney. Her laboratory studies were also all within normal parameters, and the physical examination failed to detect abnormal manifestations. The patient then underwent the total resection of the right kidney with the tumor in our hospital.

2.3. Immunohistochemal staining

The resected specimens were fixed with 10% neutral-buffered formalin, then embedded in paraffin blocks and cut into 4-μm thickness slides. The slides were deparaffinized with xylene, rehydrated with graded alcohols, and incubated using the following antibodies: cytokeratin, smooth muscle actin (SMA), S-100, epithelial membrane antigen (EMA), GLUT1, CD34, progesterone receptor (PR), HMB45, MelanA, MiTF, Desmin, CD99, STAT6, CD21, CD23, CD117, Dog-1, calponin, CD68, CD10, and Ki-67, and stained with a streptavidin–peroxidase system (KIT-9720, Ultrasensitive TM S-P, MaiXin, China). The chromogen used was diaminobenzidine tetrahydrochloride substrate (DAB kit, MaiXin, China). Appropriate positive and negative controls were used to exclude the false positivity and negativity.

2.4. Morphological and immunohistochemical findings

Grossly, there was a solid mass approximately 6.5 cm in diameter at the lower pole of the kidney and partly in the renal pelvis. The mass appeared to be relatively well circumscribed, and the capsule was absent at the periphery of the tumor. The cut face was firm and gray-yellow in color (Fig. 1).

Histologically, the tumor straightforward involved the renal parenchyma, forming the pushing border at the periphery. The tumor was predominantly composed of bland ovoid to short spindle cells with clear cytoplasm, which formed characteristic storiform pattern. Focally, a number of spindle cells were arranged in a fascicular architecture, reminiscent of cutaneous cellular FH. The cellular atypia and mitotic activities were absent. Moreover, scattered inflammatory cells were present among the cells. Overall, the blood vessels in the tumor were not remarkable, very focally, dilated and branching vessels could be observed (Fig. 2).

Immunohistochemically, the epithelial cells showed diffusely positive for vimentin, focally positive for SMA, and consistently negative for cytokeratin, S-100, EMA, PR, GLUT1, CD34, HMB45, MelanA, MiTF, Desmin, CD99, STAT6, CD21, CD23, CD117, Dog-1, calponin, CD68, and CD10. Ki-67 index was less than 5% (Fig. 3).

Taken together, according to the morphological profile and immunohistochemical staining, the tumor was diagnosed as a deep BFH.

2.5. Follow-up

The patient was alive with no tumor recurrence or metastasis within 6 months of follow-up.

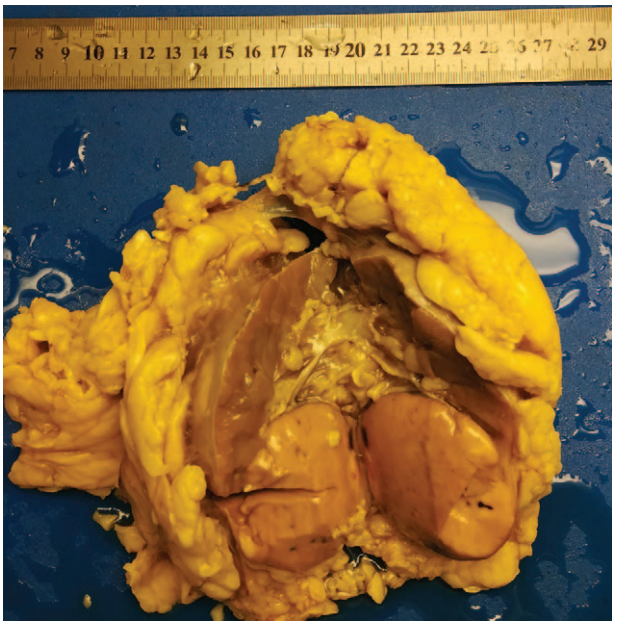

Figure 1. Macroscopic manifestation of the tumor. The lower pole of the right kidney and renal pelvis was occupied by a solid well-circumscribed mass measuring 6.5 cm in diameter with a firm and gray-yellow cut face.

3. Discussion

It is relatively uncommon for soft tissue tumors of the kidney except for angioleiomyolipoma.[13,14] Other tumors, including solitary fibrous tumor (SFT) and hemangioma, are occasionally encountered in this location.[15,16] Deep BFH of kidney is extremely rare. Thus far, only one case reportedly occurred in the kidney.[11] The present case is another that involved parenchyma of the kidney.

Superficial BFH is one of the most frequent tumors of skin, which could show variable histologic manifestations.[1–3] Instead, deep BFH usually possessed the distinct characteristics.[6] Macroscopically, similar to the present case, deep BFH appeared as a well-circumscribed mass and showed a well-defined border, in contrast to superficial BFH. Moreover, deep BFH usually was bigger than cutaneous BFH, because of its deep location. Histologically, deep BFH typically comprised bland ovoid to spindle cells arranged in a storiform pattern with admixed lymphocytes in common with cutaneous BFH. However, the storiform pattern was more prominent than cutaneous BFH. And the branching vessels or hemangiopericytoma-like structure in deep BFH were more often encountered, although the vessels were usually focally present. The inflammatory cells such as foam cells and Touton giant cells appeared to be not as often as in cutaneous BFH.[10] Our case demonstrated the similar morphology with the case reported by Sakakibara et al,[11] the ovoid cells or short spindle cells lacking atypia and mitosis formed prominent storiform structures. Foam cells and multinucleated cells were not present in both the cases.[11] Focally, dilated and branching vessels could be observed in our cases. The case reported by Kobayashi et al[12] that occurred in the renal capsule also demonstrated the similar morphologic manifestations.

It seems that the histochemical staining is not helpful for diagnosis of BFH. According to Gleason and Fletcher,[6] deep BFH mainly stained for smooth muscle actin in 15/40 (38%) and

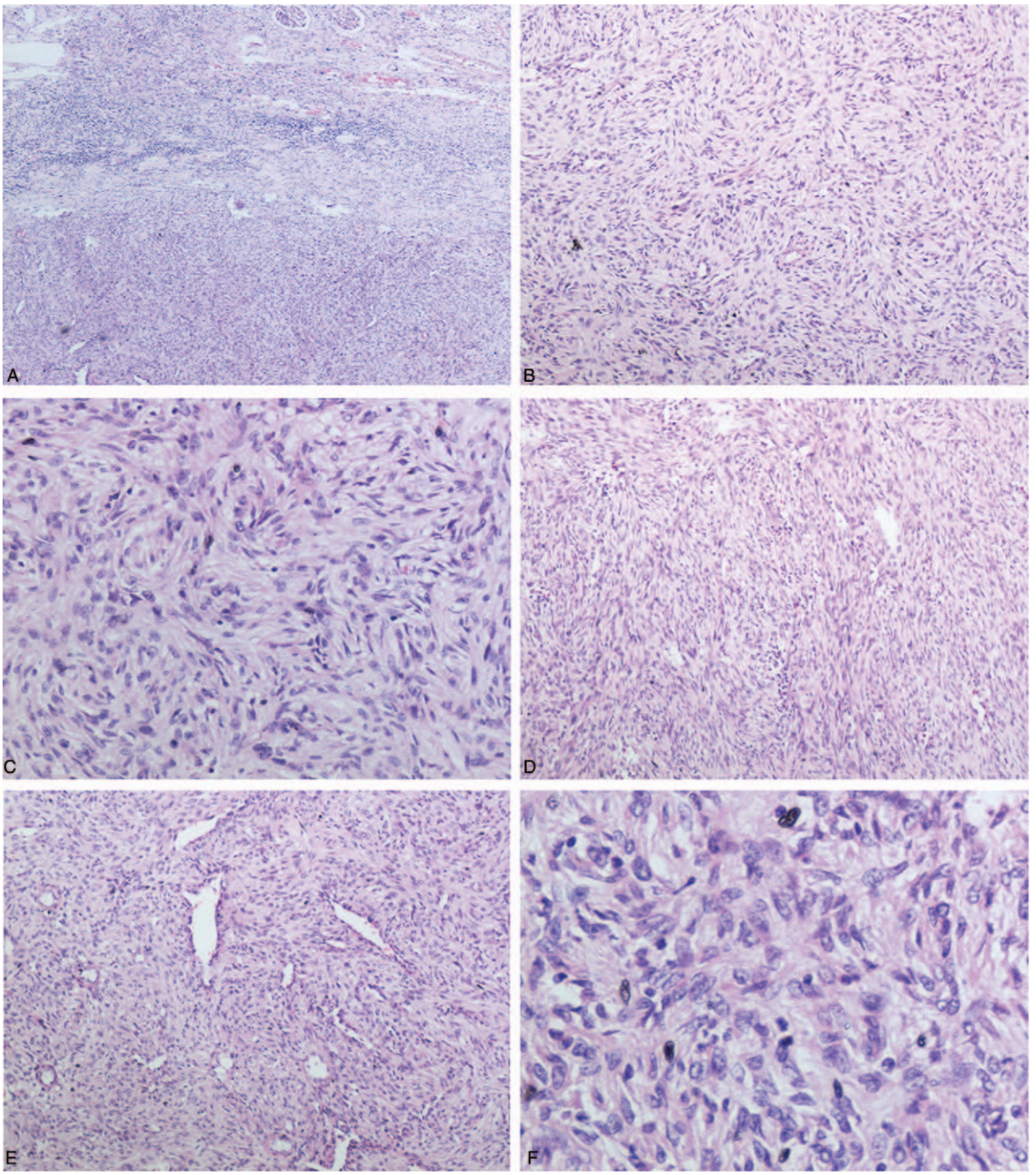

Figure 2. Morphological change of the tumor. (A) Lower magnifications revealed that the tumor was relatively well circumscribed, but lacked the fibrous capsule at the periphery. (B, C) The ovoid cells formed prominent storiform structures. (D) Focally, spindle cells were arranged in a fascicular architecture, reminiscent of cutaneous cellular FH. (E) The dilated and branching vessels could be focally observed. (F) The cells were generally ovoid and lacked atypia and mitosis. Scattered inflammatory cells were present among the cells. FH = fibrous histiocytoma.

desmin in 1/12 (8%), which were not specific for BFH.[11] It was originally thought that FH was from fibroblastic and histiocytic cells. In fact, BFH demonstrated no evidence of true histiocytic cells differentiation, as the markers CD68 and CD163 were frequently negative in the tumor.[1–4,17,18] The main aim of histochemistry is to exclude the other tumors. However, Gleason and Fletcher[6] also reported that 40% of deep BFH cases were positive for CD34,[11] which could lead to be a misdiagnosis.

The main differential diagnosis of the tumor includes SFT, perineurioma, and malignant FH (undifferentiated polymorphic sarcoma). SFT is characterized by patternless pattern rather than storiform pattern. It should be noted that deep BFH may also stain for CD34, which can also be expressed in SFT. We can add STAT6 staining, a marker recently reportedly specific for SFT, for differential diagnosis.[19] In our case, the tumor was negative for CD34, CD99, and STAT6, which could absolutely exclude SFT.

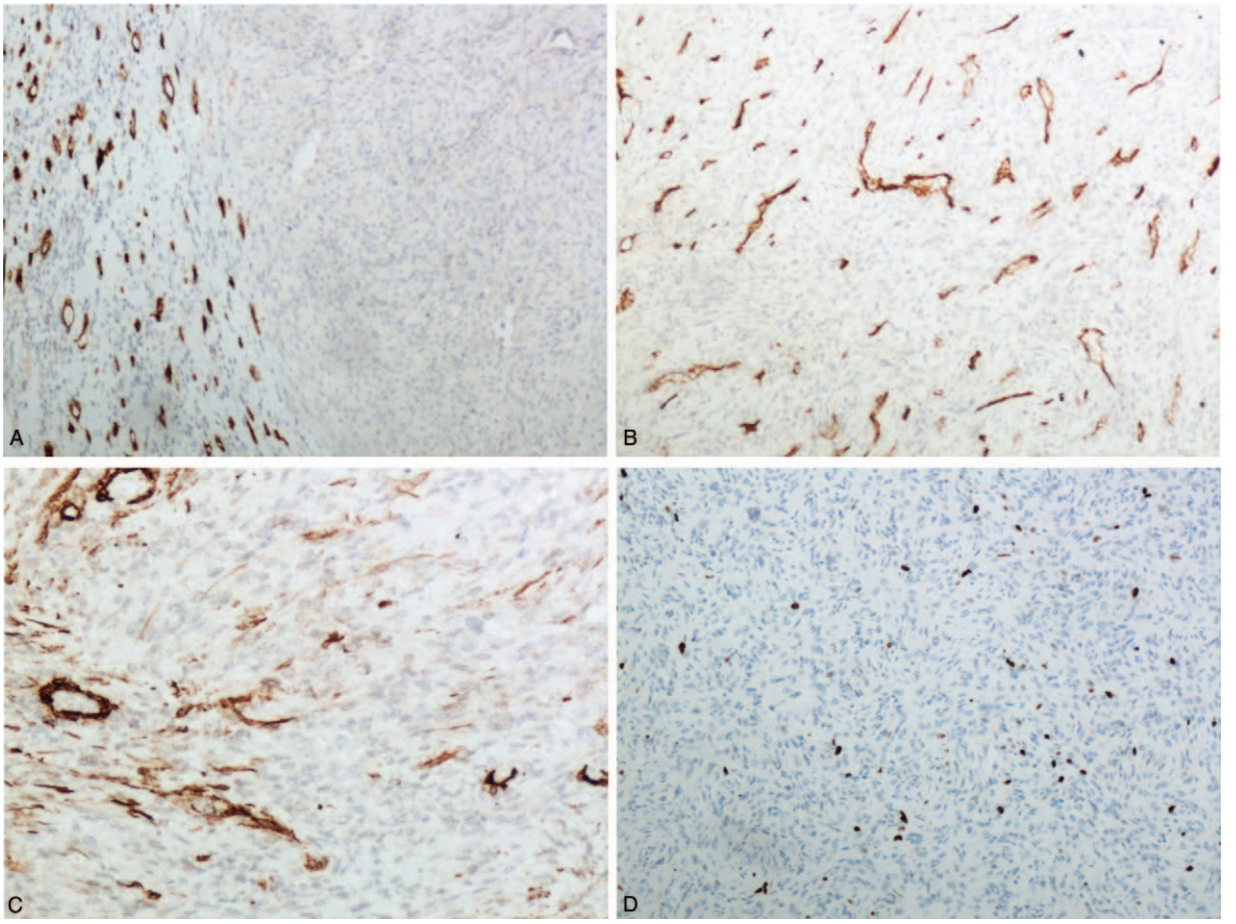

Figure 3. Immunohistochemical staining of the tumor. (A) The tumor cells were positive for EMA. (B) The CD34 staining highlighted the presence of vessels in the tumor. (C) SMA was focally expressed in the tumor cells. (D) Ki-67 proliferative index was less than 5%. EMA = epithelial membrane antigen, SMA = smooth muscle actin.

Perineurioma occasionally occurred in the kidney.[20,21] Perineurioma can also demonstrate prominent storiform structure. However, perineurioma has thin, elongated, spindled cells with slender nuclei. Immunohistochemically, perineurioma is consistently positive for EMA and GLUT1,[22] which is also helpful for differential diagnosis. Moreover, the lack of cellular atypia and mitosis can rule out malignant FH (undifferentiated polymorphic sarcoma).

Clinically, deep BFH is generally considered as a benign tumor. Nevertheless, in the limited follow-up data, deep BFH may recur and rarely metastasize.[15] In our case, as the tumor was large and straightforward involved the kidney, the complete excision of the tumor and then a longer follow-up were necessary.

Author contributions

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