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Case Report

Characteristic findings of primary perirenal angiosarcoma on chemical shift subtraction magnetic resonance imaging (CSS-MRI): A case report ☆

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ABSTRACT

A 70-year-old man with supraglottic carcinoma underwent computed tomography (CT) for staging purposes. A tumor measuring approximately 7 × 10 cm was found incidentally in the left perirenal space. The tumor showed homogeneous high signal intensity on chemical shift subtraction magnetic resonance imaging (CSS-MRI) suggesting the presence of minimal amounts of fat. Five months later, the tumor had grown to approximately 10 × 12 cm with indistinct margins. CSS-MRI showed high signal intensity in the tumor periphery only. The tumor was resected and the pathological diagnosis was angiosarcoma. Angiosarcomas are malignant endothelial vascular neoplasms that are highly invasive to their surroundings. Here we report a case of primary perirenal angiosarcoma that was difficult to differentiate from a dedifferentiated liposarcoma. On CSS-MRI, high signal intensity within a tumor may be a characteristic feature of primary perirenal angiosarcoma.

Introduction

Angiosarcoma is a rare and highly aggressive malignant tumor that arises from the endothelium of the blood or lymphatic vessels. Angiosarcomas can arise in any soft-tissue structure or visceral organ, including the breast, liver, heart, bone, and spleen [1]. Angiosarcomas arising in the perirenal space are extremely rare. To our knowledge, only two case reports of primary perirenal angiosarcoma have been documented to date [2,3]. Here we report a case of primary perirenal angiosarcoma that was difficult to differentiate from a

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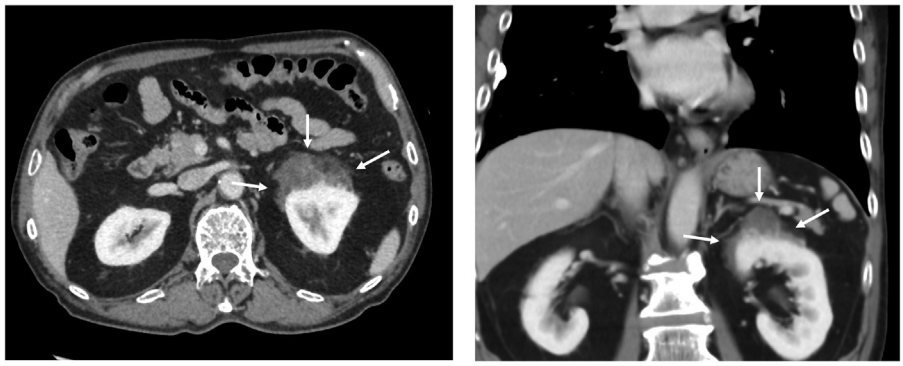

Fig. 1 – Axial (A) and coronal (B) contrast-enhanced computed tomography (CT) image showing a tumor in the left perirenal space measuring approximately 7 x 10 cm (A, B arrows). The tumor shows heterogeneous enhancement and is contiguous with the kidney on contrast-enhanced CT. The tumor periphery shows low attenuation at -5 Hounsfield units, indicating fat density.

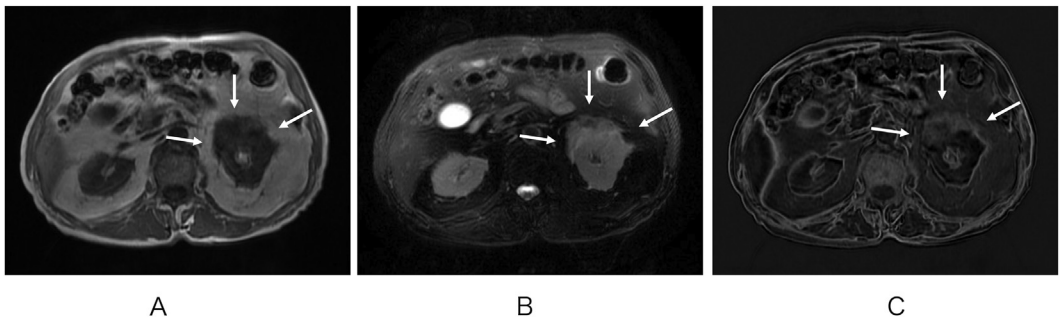

Fig. 2 – The tumor shows low signal intensity on T1-weighted image (A) and relatively homogeneous high signal intensity on fat-suppressed T2-weighted image (B). Chemical shift subtraction magnetic resonance imaging (C) shows homogeneous high signal intensity, indicating the presence of small amounts of fat throughout the tumor.

dedifferentiated liposarcoma due to the presence of minimal amounts of fat in the tumor on chemical shift subtraction magnetic resonance imaging (CSS-MRI).

Case presentation

A 70-year-old man with supraglottic carcinoma underwent computed tomography (CT) for staging purposes. A tumor measuring approximately 7 × 10 cm was found incidentally in the left perirenal space. The tumor showed heterogeneous enhancement and was contiguous with the kidney on contrast-enhanced CT. The tumor periphery showed low attenuation at -5 Hounsfield units, indicating fat density (Fig. 1). The tumor showed low signal intensity on T1-weighted imaging (Fig. 2A) and relatively homogeneous high signal intensity on fat-suppressed T2-weighted imaging (Fig. 2B). CSS-MRI showed homogeneously high signal intensity, indicating the presence of minimal amounts of fat throughout the tumor (Fig. 2C).

Five months later, the tumor had grown to approximately 10 × 12 cm with indistinct margins. The tumor showed heterogeneous high signal intensity on fat-suppressed T2-weighted imaging (Fig. 3A). CSS-MRI showed high signal intensity in the tumor periphery only (Fig. 3B). Dynamic contrast-enhanced MRI (DCE-MRI) showed gradual nodular enhancement from the periphery to the center (Fig. 3C). As the tumor was retroperitoneal and contained fat, it was preoperatively diagnosed as a dedifferentiated liposarcoma.

A radical nephrectomy including the ipsilateral adrenal gland was performed. The tumor was not demarcated from the kidney, and its cut surface was dark brown macroscopically due to blood (Fig. 4). Hematoxylin and eosin staining showed dark staining of the nucleus, the proliferation of atypical spindle-shaped cells, and pseudovascular spaces (Fig. 5A). Tumor cells were found in the perirenal fat tissue (Fig. 5B). Immunohistochemical staining showed positivity for CD31, CD34, factor VIII, and p16 (Figs. 5C and D). A histopathologic examination confirmed the diagnosis of angiosarcoma.

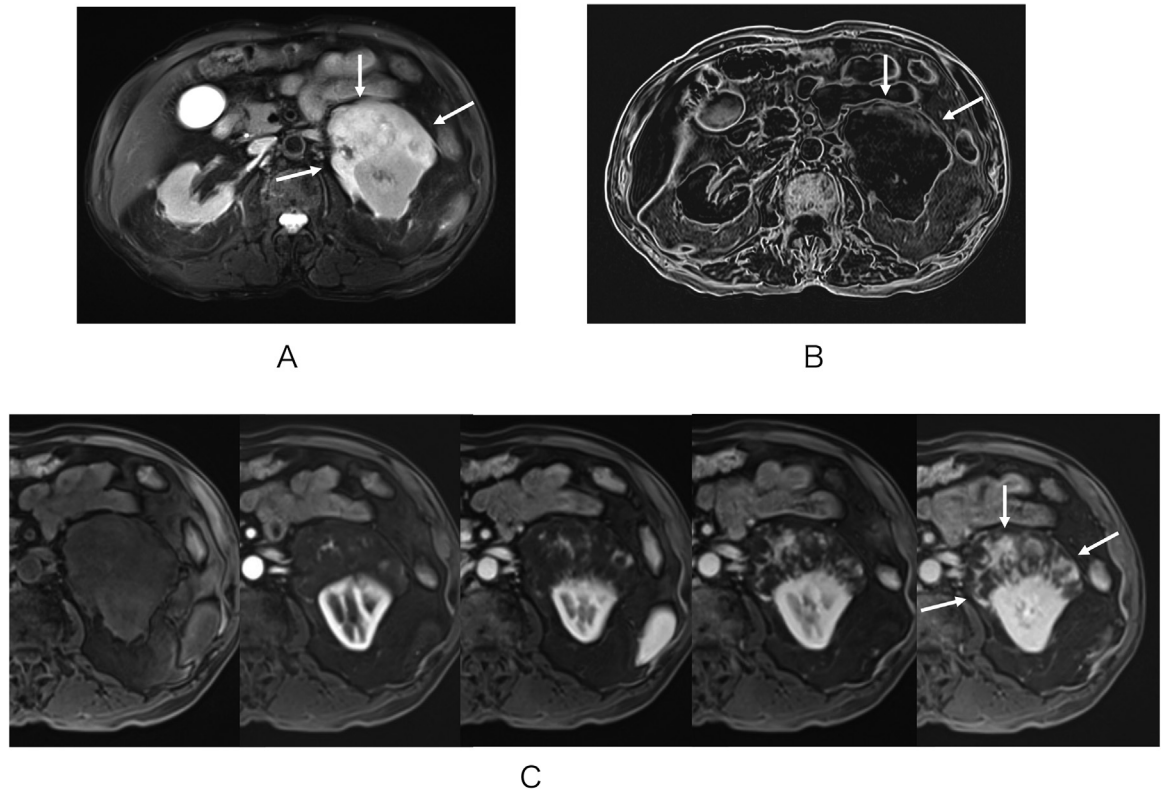

Fig. 3 – Five months later, the tumor was approximately 10 × 12 cm with ill-defined margins. The tumor shows a heterogeneous high signal intensity on fat-suppressed T2-weighted image (A). Chemical shift subtraction magnetic resonance imaging (B) shows high signal intensity at the tumor periphery only. Dynamic contrast-enhanced magnetic resonance imaging (C) shows gradual nodular enhancement from the periphery to the center.

Fig. 4 – The angiosarcoma is not demarcated from the kidney, and the cut surface was dark brown macroscopically due to blood (arrows).

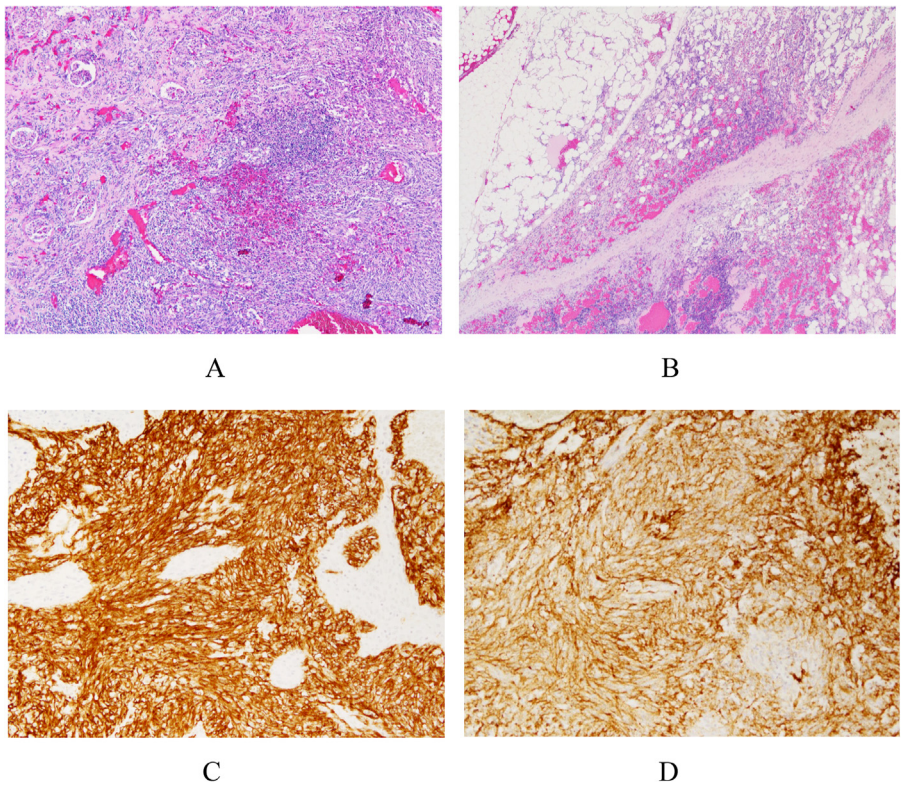

Fig. 5 – Hematoxylin and eosin staining revealing a dense proliferation of atypical spindle-shaped cells with darkly stained nuclei and pseudovascular spaces (A). The infiltration of tumor cells into the perirenal fat tissue is visible (B). Immunostaining tests are positive for CD34 (C) and factor VIII (D).

An abdominal CT performed at 12 months postoperative showed local recurrence and multiple metastatic liver tumors. The patient was treated with molecularly targeted drugs, but the treatment was ineffective and his condition worsened. He ultimately died at 24 months postoperative.

Discussion

Angiosarcomas are malignant endothelial vascular neoplasms that invade the surrounding structures. On pathological examination, the tumor cells invaded the surrounding adipose tissue. Over time in this case, on CSS-MRI, a change was noted from high signal intensity across the entire tumor to the tumor periphery only. Therefore, high signal intensity in the tumor on CSS-MRI may be a characteristic feature of primary perirenal angiosarcoma with perirenal adipose tissue invasion.

Dedifferentiated liposarcomas represent a biphasic neoplasm, with one component being a well-differentiated liposarcoma and the other a non-adipose cellular sarcoma [4]. However, the components of the well-differentiated liposarcoma are occasionally undetectable on conventional MRI. Kransdorf et al.[5] reported that chemical shift imaging is useful for depicting microscopic fat in high-grade liposarcoma. In retroperitoneal tumors, soft-tissue tumors with fatty components are most likely dedifferentiated liposarcomas. Our case was misdiagnosed as a dedifferentiated liposarcoma prior to surgery because we thought that the microscopic fat detected on CSS-MRI was derived from the tumor. Pathologically, the microscopic fat observed at the tumor's periphery was due to the involvement of adipose tissue around the angiosarcoma. In this way, the fact that the fat components were not derived from the tumor distinguished it from dedifferentiated liposarcoma.

In our case, DCE-MRI showed gradual nodular contrast enhancement from the periphery to the center. These imaging findings are consistent with those of previous studies of angiosarcomas arising from the liver [6,7]. The histology of angiosarcomas does not differ according to the site of origin, with an increase in irregularly dilated and anastomosing lumens and proliferation of atypical and polymorphous endothelial cells that spread to divide the fibrous stroma and matrix [1]. Gradual nodular contrast enhancement on DCE-MRI may indicate an increased number of dilated and anastomosing lumens. Therefore, a combination of gradual nodular contrast enhancement on DCE-MRI and high signal intensity in the tumor on CSS-MRI would place primary perirenal angiosarcoma at the top of the list of differential diagnoses.

Conclusion

Here we reported a case of primary perirenal angiosarcoma with perirenal fat tissue invasion. Our findings suggest that

high signal intensity within a tumor on CSS-MRI may be a characteristic feature of primary perirenal angiosarcoma.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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