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Case Report

Retroperitoneal dedifferentiated liposarcoma with rare heterologous low-grade osteosarcoma: A case report ☆

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ABSTRACT

Dedifferentiated liposarcoma is a high-grade entity developed from a preexisting or recurrent well-differentiated liposarcoma, and rarely, it may contain divergent differentiation. We presented the case of a 39-year-old woman with retroperitoneal dedifferentiated liposarcoma with heterologous low-grade osteosarcoma, possessing a special pattern of tumoral calcification.

Background

Liposarcoma stands out as the most prevalent primary retroperitoneal sarcoma, constituting approximately 35% of all malignant soft-tissue tumors found in the adult retroperitoneum. These tumors are characterized by their imaging features, appearing as encapsulated fatty masses causing mass effects on adjacent structures. There are 4 subtypes: well-differentiated liposarcoma (WDL), myxoid liposarcoma, pleomorphic liposarcoma, and dedifferentiated liposarcoma (DDL). Among these subtypes, well-differentiated liposarcoma

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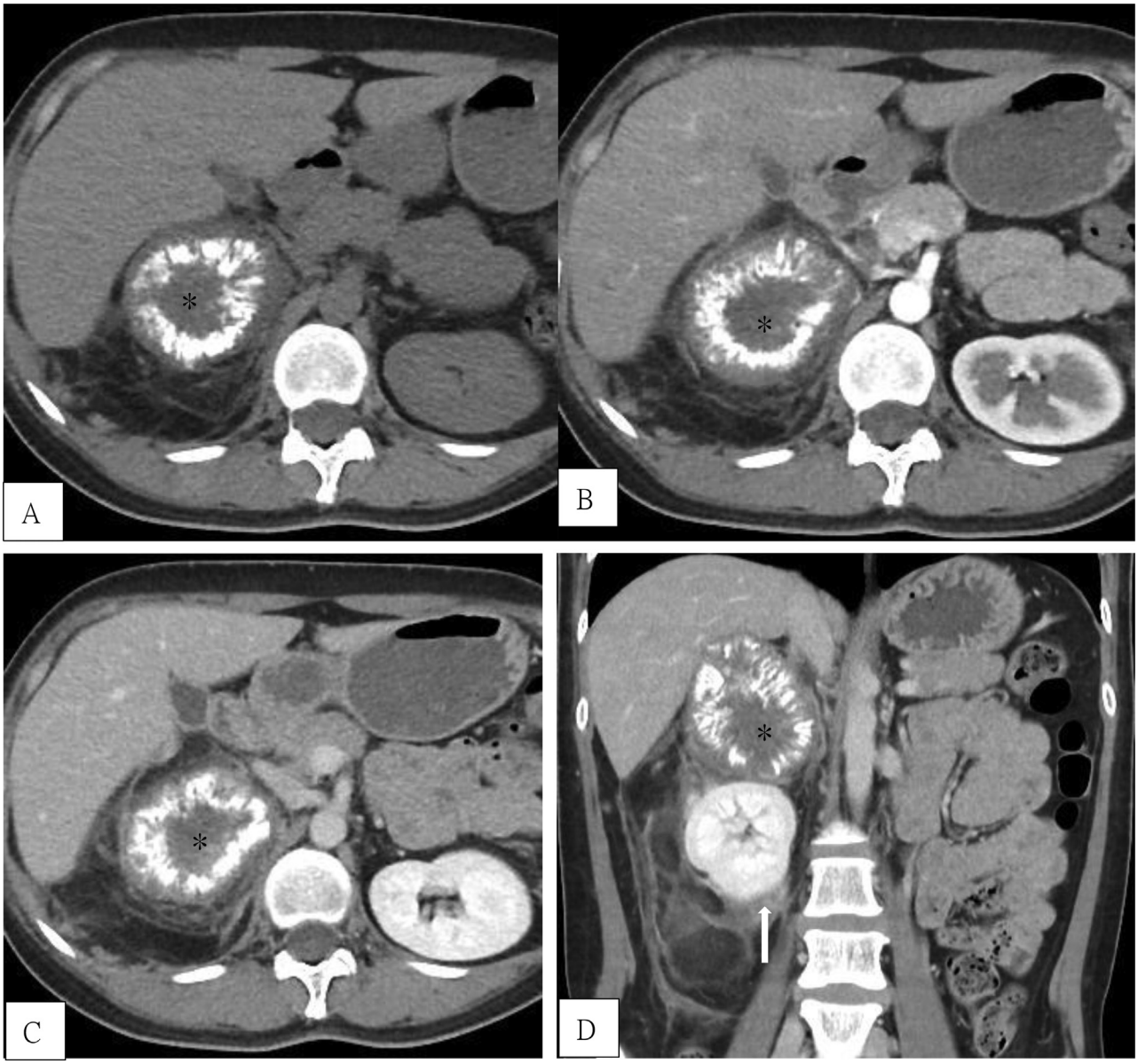

Fig. 1 – Dynamic abdominal CT, axial scan of noncontrast phase (A), axial scan of corticomedullary phase (B), axial scan of nephrographic phase (C) and coronal reconstruction images of nephrographic phase (D), demonstrate a right suprarenal solid mass with internal numerous curvilinear radiated calcifications (asterisks in A–D) in a sunburst pattern. Soft-tissue content was found inside the fatty component (arrow in D).

takes the lead as the most frequently encountered, with dedifferentiated liposarcoma following closely behind [1].

Dedifferentiated liposarcoma, classified as high-grade sarcoma by the World Health Organization (WHO) classification system, is thought to developed de novo inside preexisting WDL or in a recurrent WDL [1]. Histologically, dedifferentiated areas inside DDLs typically exhibit a higher proportion of more soft-tissue components, often resembling undifferentiated pleomorphic sarcoma or myxofibrosarcoma [2]. Rarely, divergent differentiation, including the development of rhabdomyosarcomatous, leiomyosarcomatous, or osteosarcomatous components, has been reported in less than 5% of DDLs [2,3].

Hereby we present a case of primary retroperitoneal DDL with heterologous osteosarcoma.

Case presentation

A 39-year-old woman without a known cancer history presented with sudden-onset epigastric pain for a month. She went to the emergency department nearby, where her abdominal sonography found a 4 cm mass near her right kidney. Further abdominal computerized tomography (CT) disclosed a 6 cm calcified mass with a peripheral adipose com-

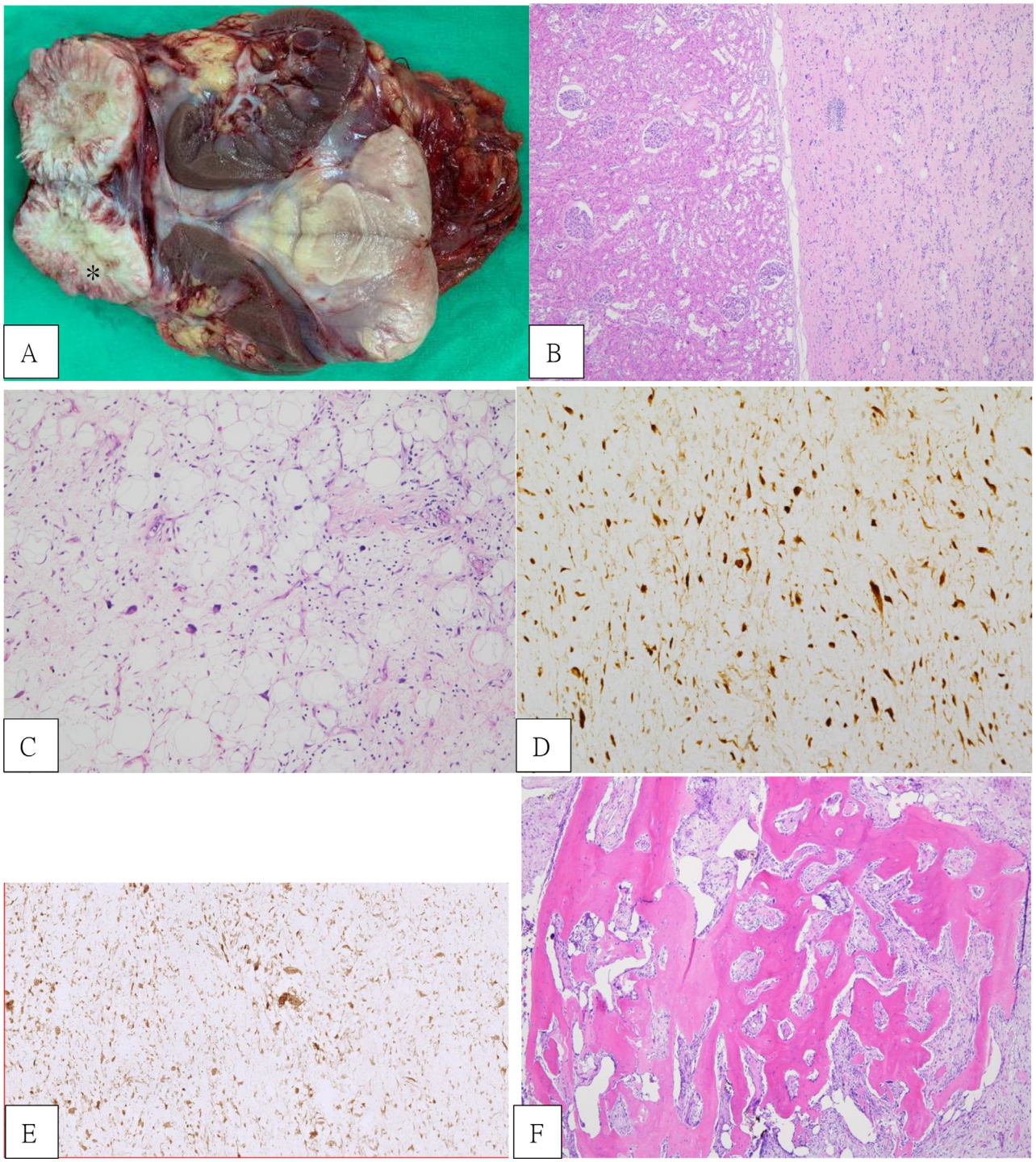

Fig. 2 – Gross pathological specimens (A) reveal a fatty mass encasing the kidney and a bony nodule (asterisk in A). A microscopic exam of the tumor section showed dedifferentiated liposarcoma encasing the kidney (B), which also contained a low-grade osteosarcoma component (C). Immunohistochemistry disclosed CKD4 (D) and MDM2 (E) positive. (F) The section of the bony nodule showed heterologous low-grade osteosarcoma.

ponent at the right suprarenal space and an extending component encased the right kidney. Surgical removal of the tumor with nephrectomy was suggested there. The patient hesitated and 4 months later she went to our outpatient department of urologist for a second opinion. Abdominal CT with dynamic phases was performed again, and showed a suprarenal solid mass with internal numerous curvilinear radiated calcifications in a sunburst pattern (Fig. 1). It enlarged to 7.4 cm, downward displacing the right kidney. Also, the peripheral adipose component encased the right kidney with soft-tissue

content inside the adipose component. There was no right kidney hydronephrosis, adjacent enlarged lymphadenopathy, or right renal vein thrombosis. Neither did we find evident lung or bone metastasis in available images. Given the absence of contraindications, surgical removal was arranged and total removal with right nephrectomy and adrenalectomy were done.

The macroscopic pathological specimens were 19.4 × 14.5 × 8.4 cm in size, containing a kidney, an adrenal gland, and a firm bony nodule 8.2 × 6.9 × 6.7 cm in size (Fig. 2). The tumor involves perirenal and periadrenal fat, but does not invade the adrenal gland, ureter, or hilar vessels. Sections of the tumor showed liposarcoma, which was composed of both well-differentiated and dedifferentiated components with heterologous low-grade osteosarcoma. The tumor cells were immunoreactive for CDK4 and MDM2. Diagnosis of dedifferentiated liposarcoma with heterologous low-grade osteosarcoma was made.

After surgical treatment, the patient received local radiotherapy of 6300 cGy in 30 fractions. Subsequently, the patient has been monitored through regular follow-up appointments at our urology department. As of the drafting of this case report, approximately 5 years postsurgery, there has been no evidence of recurrence.

Discussion and conclusions

DDL is a high-grade disease and mostly arises from retroperitoneum, less frequently in inguinal area and extremities and is associated with high rates of local and metastatic recurrence. It commonly occurs in the fifth to seventh decades of life and without sexual differences [1,4,5]. About 90% of DDLs develop within primary WDLs, and 10% from local recurrent WDLs [4]. The most common dedifferentiated component is undifferentiated pleomorphic sarcoma (previously called malignant fibrous histiocytoma) or fibrosarcoma. Divergent differentiation of DDLs, including the myogenic, angio- sarcomatous, and/or osteochondromatous components, are rare and have been reported in 5%-10% of DDLs [6–9].

The image feature of DDLs, consistent with its' origin from WDLs, is bimorphic neoplasm with well-demarcated fatty mass and non-fatty dedifferentiated soft tissue components, which can be readily identified on CT and MR [1,9]. Calcification or ossification is a distinct imaging pattern of DDL, which can be a result of divergent osteosarcomatous or chodrosarcomatous differentiation [10]. And according to Tateishi et al. [10], although the sample was small (3 of 6 DDLs with calcified metaplasia), univariate analysis revealed that the calcification or ossification had a significant impact on overall survival.

The patterns of calcification or ossification in DDLs vary from one case to another. Tateishi et al. reported a patient with small spotty calcifications inside DDL [10]. Yoshida et al. [11] described a case characterized by well-differentiated liposarcoma containing low-grade osteosarcoma containing internal dense bulky calcification. Fujii et al. [2] presented a recurrent retroperitoneal DDL with high-grade osteosarcomatous transformation, with about 50% dense calcification inside a solid soft-tissue mass. The tumor of our case exhibited numerous curvilinear radiated calcifications, somehow mimicking the sunburst pattern of primary osteosarcomas at extremities.

When making a diagnosis of an osteosarcoma, immunochemistry exams have crucial roles in differentiation and determining tumor grade. Dujardin et al. [12] promoted that immunohistochemical expression of MDM2 and CDK4 helps differentiate low-grade osteosarcoma from other benign fibrous and fibro-osseous lesions. Also, Yoshida et al. [13] found that MDM2/CDK4 immunohistochemical co-expression is sensitive and specific in high-grade osteosarcomas that progressed from low-grade osteosarcomas, while primary and recurrent/metastatic high-grade osteosarcomas rarely present MDM2 and CDK4 co-expression. However, it's worth noting that DDLs also express MDM2 and CDK4. In our case, pathological diagnosis of osteosarcoma was primarily based on morphology and immunochemistry might have a limited role in our case.

Nevertheless, radiologists should exercise caution when making diagnosis of retroperitoneal DDL with calcification. Renal angiomyolipoma is a common benign entity when considering differential diagnosis for retroperitoneal fatty tumors with calcification. Wang et al. [14] summarized useful practicing points for differentiated renal angiomyolipoma from retroperitoneal DDL with calcification, including renal parenchymal defect at the site of tumor contact, dilated intratumoral vessels, and the presence of renal parenchymal vascular pedicle.

Conclusion

In conclusion, we presented a rare case of retroperitoneal dedifferentiated liposarcoma with heterologous low-grade osteosarcoma. Radiologists should be cautious when dealing with retroperitoneal fatty mass with calcification.

Patient consent

Written informed consent was obtained from the patient's legal representative for the publication.

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