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CASE REPORT

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Low-grade liposarcoma with osteosarcomatous dedifferentiation: radiological and histological features

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Abstract We describe the radiological and pathological findings of a rare case of a low-grade liposarcoma associated with a high-grade osteosarcomatous component in a 78-year-old woman. Pre-operative imaging demonstrated a well-encapsulated homogeneous fatty tumour in the right buttock deep to the gluteal musculature. Centrally within the fatty tumour a region of increased soft tissue attenuation on CT and heterogeneous high signal on fat-saturated T2-weighted MRI was seen with amorphous calcification/ossification. Histological assessment revealed a low-grade liposarcoma containing an area of dedifferentiation that resembled an osteosarcoma. We describe the radiological-pathological findings in this rarely reported clinical entity and discuss the differential diagnosis of calcification within lipomatous tumours.

Keywords Liposarcoma · Osteosarcoma · Dedifferentiation · CT · MRI

Introduction

It is well recognised that dedifferentiated non-lipomatous sarcomas can be found in low-grade liposarcomas [1, 2, 3, 4, 5, 6, 7, 8, 9, 10]. These arise from a focus of low-grade tumour which dedifferentiates to a tumour of a higher grade. They can also occur de novo as a component of the original tumour. Dedifferentiated elements are most commonly malignant fibrous histiocytomas, with other histological types occurring less frequently [3, 6, 7]. Dedifferentiated osteosarcomatous elements within lipomatous lesions occur rarely, with only four cases reported in the pathological world literature [1, 4, 7, 10]. We present the radiological and pathological findings of this rare entity and discuss the differential diagnosis of calcification/ossification within lipomatous tumours.

Case report

A 78-year-old woman was referred to our institution because of a gradually enlarging mass in the right buttock. On physical examination the right buttock was larger than the left but the mass was difficult to define by palpation and was non-tender. The mass had been imaged 18 months previously by CT of the abdomen and pelvis performed at an outside institution to stage a colonic carcinoma. The mass was demonstrated to be of fat attenuation with a central focus of calcification/ossification. Based on these imaging findings the lesion was felt to be either a lipoma or well-differentiated low-grade liposarcoma with a focus of central dystrophic calcification/ossification. After an uneventful recovery from a subtotal colectomy the patient underwent a repeat CT 16 months after the first CT examination. The mass was noted to be enlarging and the patient was referred for an oncological assessment.

CT performed at the time of referral to our institution (contiguous helical axial slices with collimation of 5 mm, pitch 1.0, reconstructed every 2.5 mm; Siemens Somatom Volume Zoom) through

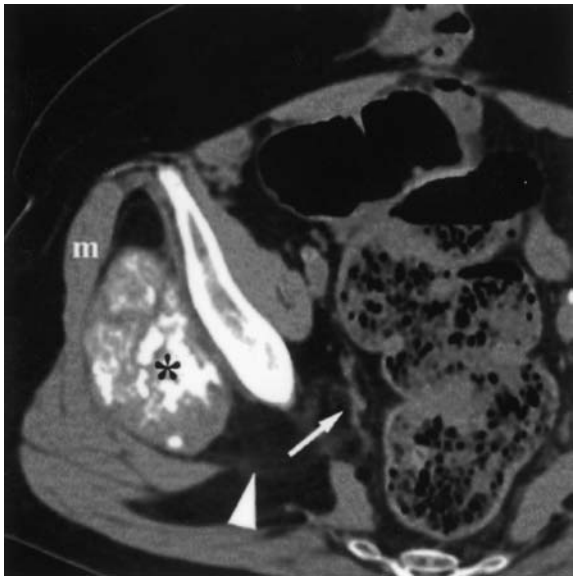

Fig. 1 A 5 mm axial CT scan of the pelvis demonstrating a well-encapsulated fat attenuation tumour deep to gluteus minimus and tensor fascia lata (*m*) extending medially through the greater sciatic notch (*arrow*). Within the fatty tumour there is a region of non-lipomatous soft tissue attenuation containing multiple amorphous areas of calcification/ossification (*asterisk*), which is associated with fatty stranding of the host tumour (*arrowhead*)

the pelvis and proximal thigh without intravenous contrast medium revealed a lobulated mass measuring approximately 20×14×5 cm. The mass lay deep to the gluteus medius muscle and extended inferiorly into the posterolateral aspect of the proximal right thigh and herniated medially through the greater sciatic notch displacing the sciatic nerve medially (Fig. 1). The mass abutted the adjacent iliac bone but there was no evidence of cortical destruction. The mass was of fat attenuation with a central well-defined oval mass of soft tissue attenuation measuring 5×8×10 cm containing dense well-defined central amorphous calcification/ossification with more diffuse peripheral mineralisation. The presence of the soft tissue component excluded the previous diagnosis of fat necrosis or dystrophic calcification within a lipoma. The distribution of calcification/ossification within the soft tissue component suggested dedifferentiation of a low-grade liposarcoma to an osteosarcoma.

MR imaging (7 mm contiguous axial and coronal T1-weighted spin echo and fat-saturated T2-weighted fast spin echo; 1.5 T, General Electric) was performed of the right pelvis and upper thigh. These images demonstrated a lesion of predominantly fat signal intensity within the anatomical location described above. A large central heterogeneous ovoid soft tissue component of the lipomatous mass was present which was hypointense to muscle on T1-weighted images (Fig. 2A) and hyperintense on T2-weighted images (Fig. 2B). Centrally within the soft tissue component there were punctate hypointense foci corresponding to the areas of calcification on CT.

A CT-guided core biopsy was performed which was interpreted histologically as extraskeletal osteosarcoma. The tumour was completely excised en bloc via a postero-lateral approach reflecting both gluteus maximus and medius and dissecting the tumour off the sciatic nerve.

Histologically the tumour consisted of two components. There was well-differentiated lipoma-like liposarcoma consisting of adipocytes with vacuoles of varying size (Fig. 3A). Admixed scattered occasional lipoblasts and cells with nuclear atypia were present. In addition there were nodules containing cells with nuclear atypia and which in areas produced osteoid which showed focal calcification (Fig. 3B). In these nodules neither adipocytes nor lipoblasts were seen.

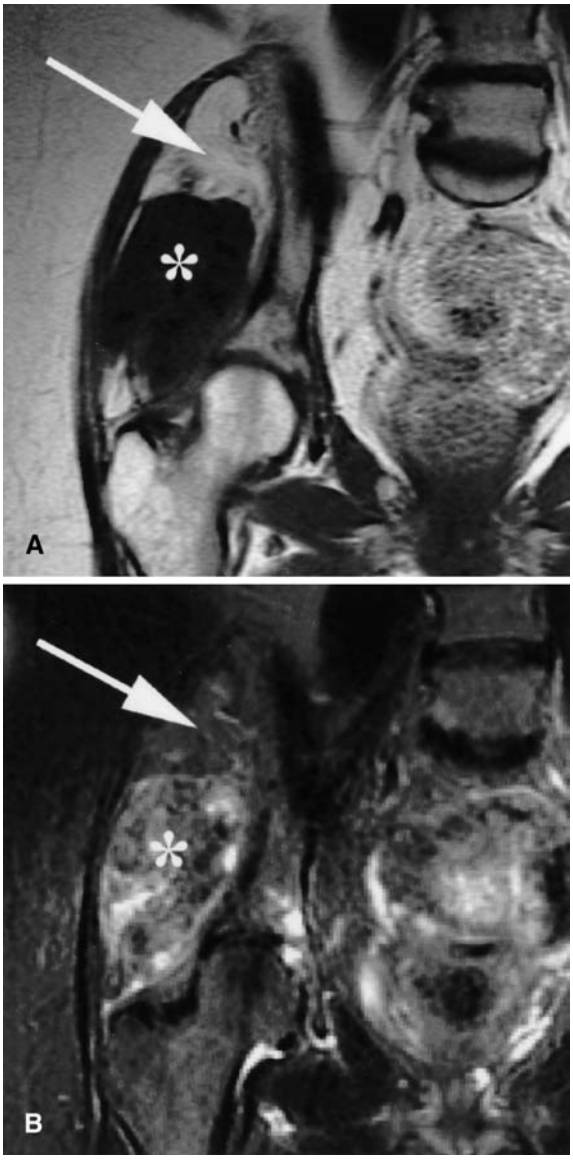

Fig. 2 **A** Coronal T1-weighted MR image of the right hip. The peripheral regions of the tumour (*arrow*) are of homogeneous high signal intensity, consistent with fat, whereas the central tumour (*asterisk*) is of heterogeneous intermediate and low signal intensity. **B** Fat-saturated coronal T2-weighted images confirm the fatty nature of the peripheral tumour by nulling the signal return (*arrow*). The central tumour (*asterisk*) was of heterogeneous high, intermediate and low signal intensity

The patient was most recently seen at clinical follow-up 9 months post-operatively. At this time the patient is doing well with no clinical or radiologic evidence of recurrent or residual local disease. However, follow-up CT scan of the chest has illustrated the development of new small lower lung nodules with an appearance suspicious for metastatic disease.

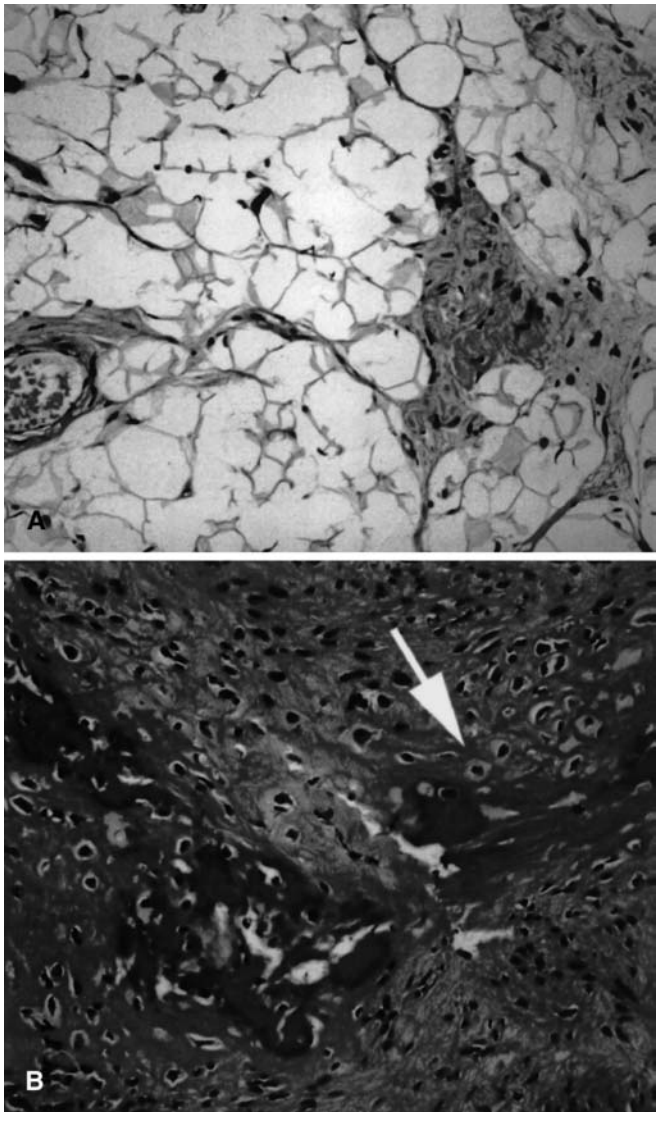

Fig. 3 Photomicrographs (hematoxylin and eosin, ×200 original magnification) of lipoma-like liposarcoma (**A**) and osteosarcoma components (**B**). Calcification of osteoid is demonstrated in **B** (*arrow*)

Discussion

Dedifferentiated elements within a well-differentiated liposarcoma can be seen as part of the spectrum of histological elements that comprise the original tumour. They can also arise secondarily in tumours, without prior evidence of dedifferentiation, and usually show a histological progression from low-grade well-differentiated lipomatous tumour to a less well differentiated non-lipomatous tumour of a higher grade [1, 7]. Half of all well-differentiated liposarcomas occur in the deep soft tissues of the extremity and one quarter of cases occur in the retroperitoneum [3]. Occurrence at other sites including the groin is less frequent. The mean time to dedifferentiation is 7–9 years following diagnosis of the well-differentiated liposarcoma [3, 7]. Dedifferentiated elements within well-differentiated liposarcomas can occur at any of these sites but have been reported most commonly in the retroperitoneum [1, 3, 7]. The incidence of dedifferentiation of well-differentiated lipomatous tumours is likely to be proportional to the length of time the tumours remain in situ rather than behavioural differences specific to tumours arising from different anatomical locations. As retroperitoneal lipomatous tumours remain occult for longer and are less amenable to surgery than those arising in the extremities, they have a longer natural history in which to dedifferentiate [3, 7]. As in this case report the "host" tumour is typically a lipoma-like liposarcoma composed mainly of mature fat cells interspersed with occasional lipoblasts [7]. Dedifferentiated tumours most commonly resemble high-grade malignant fibrous histiocytoma or fibrosarcoma [3, 6, 7, 8]. Other tissue types resembling rhabdomyosarcoma [2], leiomyosarcoma [5] and hemangiopericytoma [6] occur less frequently. Dedifferentiation into osteosarcoma is very rare with, to our knowledge, only four cases described in the world literature [1, 4, 7, 10]. Foci of osseous and chondro-osseous metaplasia have also been described [6, 11] within low-grade liposarcomas.

Calcification/ossification is a documented feature of lipomatous tumours on radiographs [12]. It is most commonly seen as a result of fat necrosis, due to trauma or ischaemia. Calcification/ossification can also occur following osseous or chondro-osseous metaplasia or, as in our patient, in rare cases of osteosarcomatous dedifferentiation. In our case the imaging demonstrated a region characteristic of non-lipomatous soft tissue on both CT and MRI within an otherwise homogeneous lipomatous lesion, which raised the possibility of a dedifferentiated liposarcoma. Within the central soft tissue mass there was ill-defined and diffuse calcification/ossification peripherally with dense mature ossification centrally. This distribution of immature to mature mineralization is similar to that described in parosteal osteosarcomas [13, 14, 15]. This is in contrast to the zonal distribution of mineralisation seen in myositis ossificans: central non-ossified foci progressing to osteoid more peripherally and dense mature ossification at the periphery [16, 17]. Ossification has been described in rare cases of chondro-osseous metaplasia and benign chondro-osseous differentiation of lipomatous tumours [6, 11]. In these cases the imaging characteristics of the ossification varied from diffuse to well defined but in all cases the distribution was predominantly peripheral and without the zonal distribution seen in our case [6, 11].

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