Case report:

Atypical lipomatous tumor with osteoid metaplasia – a rare occurrence

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Abstract:
The presence of metaplastic ossification within a atypical lipomatous tumour /well differentiated Liposarcoma (ALT/WDLPS) is a rare occurrence. We report a case ALT/WDLPS with unusual extensive metaplastic bone formation in a 30 years female who complained of swelling in the right shoulder for the past one and a half years.

Keywords: Atypical lipomatous tumor; Osteoid; Metaplasia

Introduction
Atypical Lipomatous Tumor (ALT) is a malignant tumor comprising of fat and variable number of spindle cellsw ith hyperchromatic nuclei and multi vacuolated lipoblasts. The presence of metaplastic ossification within an ALT is a rare occurrence.1 We report a case of ALT with unusual extensive metaplastic bone formation.

Case summary
A 30 year old female presented with swelling in the right upper arm for the past one and a half years. Magnetic resonance imaging revealed an altered signal intensity mass lesion measuring 71x39x34 mm located subcutaneously over the lateral aspect of right upper arm. Fine needle aspiration cytology was performed however it yielded scanty material and was inconclusive. Excision of the mass was done.

We received an encapsulated soft tissue mass measuring 7x3.9x3.4cm. Cut surface was lobulated and soft in consistency, with one area showing bony hard tissue. (Figure 1) Multiple sections taken from the mass showed sheets of adipose tissue with variation in adipocyte size and shape (Figure 2), along with occasional hyperchromatic spindle cells and lipoblasts. (Figure 3) An extensive area of ossification with osteoblastic rimming of the bony trabeculae was seen. (Figure 4)Thus, possibility of a lipomatous lesion with osseous differentiation was suggested.

Differential diagnosis included lipoma with central calcified fat necrosis, osteolipoma, chondrolipoma, pleomorphic lipoma, liposarcoma with osteoid differentiation and liposarcoma with osteosarcomatous differentiation. However, presence of lipoblasts ruled out a benign diagnosis. Further, absence of cartilage and presence of variably sized adipocytes with atypia ruled out chondroidoma and osteolipoma respectively. Necrosis, inflammatory cells and foam cells seen in calcified fat necrosis were absent. The characteristic multinucleated floret cells seen in pleomorphic lipoma were not seen. There was no evidence of giant cells/increased mitotic activity/necrosis, any myxoid stroma/chicken wire blood vessels or dedifferentiation ruling out pleomorphic liposarcoma, myxoid liposarcoma and dedifferentiated liposarcoma. Liposarcoma with osteosarcomatous differentiation was excluded on the basis of absence of any malignant osteoid. Therefore, a final diagnosis of atypical lipomatous tumor with osteoid metaplasia was made.

Discussion
Atypical lipomatous tumour (ALT) and well differentiated liposarcoma (WDL) are two names for the same entity which are histologically and genetically identical. The distinction is solely anatomic with tumors presenting in the extremities and subcutaneous tissues being labeled ALT and those in the mesentry, retroperitoneum, mediastinum,
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Figure 1: Soft tissue mass with a lobulated cut surface with one area showing bony hard tissue.

Figure 2: Sheets of adipose tissue with variation in adipocyte size and shape. [H&E, 100x]

Figure 3: Numerous hyperchromatic spindle cells and an occasional lipoblast in the centre. [H&E, 400x]

Figure 4: Osteoid with osteoblastic rimming. [H&E, 400x]

spermatic cord being called as WDL. The so called ALT have higher rates of complete surgical excision, lower rate of local recurrence and lower risk of undergoing dedifferentiation as compared to WDL. On cytogenetic analysis, ALT is confirmed by ring and giant marker chromosomes composed of amplified material from the long arm of chromosome 12, and florescence in situ hybridization, however this was not done in our case.

Ossification can be seen in many adipocytic lesions, therefore, its presence can be a diagnostic dilemma to the pathologist and radiologist. Entities like calcified fat necrosis, osteolipoma, chondrolipoma, pleomorphic lipoma, liposarcoma with osteosarcomatous differentiation should be ruled out before giving a diagnosis of ALT with osteoid metaplasia. Further, ALT should be differentiated from the other subtypes of liposarcoma by carefully appreciating the histopathological features and taking additional sections if necessary.

Osteoid metaplasia in atypical lipomatous tumor has seldom been documented in the available literature.
Gupta et al reported a mesenteric well differentiated liposarcoma with focal osseous metaplasia. Javery et al reported extensive metaplastic ossification in an ALT situated near the right shoulder. Osteoid metaplasia in malignant adipocytic lesions is usually seen in association with a dedifferentiated component arising within a primary liposarcoma. However, there was no dedifferentiation seen in our case. ALT with metaplastic ossification should be considered in the differential diagnosis for a calcified soft tissue mass. Proper diagnosis is essential for optimal surgical planning and subsequent treatment. Ethical clearance: Ethical clearance was taken prior to submission.

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Conflicts of interest: None

Author contributions:
Manjula Jain: Reported the case, edited the manuscript
Shivali Sehgal: Performed the grossing, staining, wrote the manuscript
OP Pathania: Performed the surgery

The manuscript has been read and approved by all the authors. The requirements for authorship have been met. Each author believes that the manuscript represents honest work.

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