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Inflammatory Atypical Lipomatous Tumour: A Case Report

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ABSTRACT

Liposarcomas are common soft tissue sarcomas arising predominantly in deep soft tissue and the retroperitoneum. Inflammatory variant of well differentiated liposarcoma (inflammatory atypical lipomatous tumor) is a rare type of low grade liposarcoma, occurring most often in the retroperitoneum. We are presenting a case of inflammatory atypical lipomatous tumor in a 54 year old male patient who presented with a soft tissue mass in the thigh. The tumor was encapsulated measuring about 8 * 6*4 cms. Histopathology revealed picture consistent with inflammatory atypical lipomatous tumor. Immunohistochemistry showed S-100 positivity in lipoblasts and Vimentin positivity in the stromal cells and was negative for Smooth muscle actin.

Keywords: Liposarcoma, Atypical lipomatous tumor, S-100

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INTRODUCTION

Liposarcoma is the most common soft tissue sarcoma occuring in adults. Liposarcomas are large tumors occuring most frequently in the lower extremities, retroperitoneal, perirenal, and mesenteric region; and shoulder area. Enzinger and Winslow divided liposarcoma into four types; myxoid, round cell, well differentiated, and pleomorphic[1]. Atypical lipomatous tumour (ALT)/ well differentiated liposarcoma is further sub-divided into adipocytic (lipoma like), sclerosing, inflammatory and spindle cell type. ALTs account for about 40-45% of all liposarcomas. They are common in middle aged adults showing peak incidence in the sixth decade. The common sites of ALT are the deep soft tissue of the limbs, especially thigh, followed by the retroperitoneum, the paratesticular area and the mediastinum. Clinically ALT usually are painless and presents as a deep-seated, enlarging mass that can slowly attain a very large size, particularly in the retroperitoneum. Inflammatory variant of atypical lipomatous tumor is a rare tumor, occurring most often in the retroperitoneum. In inflammatory atypical lipomatous tumor there is a predominant population of chronic inflammatory infiltrate that may at times obscure the adipocytic nature of the neoplasm[2]. We are presenting a case of inflammatory variant of atypical lipomatous tumor in the thigh.

CASE HISTORY:

A 54 year old male patient presented with a painless slowly growing soft tissue mass in the right thigh for last 15 years, with a recent increase in the size of the mass since 1 year and pain for last 3 months.

On gross examination the tumor was encapsulated measuring about 8 * 6*4 cms. Cut-section showed solid grayish white mass with areas of haemorrhage and necrosis. Microscopic examination revealed mature adipocytes of variable size and shape intermixed with spindle cells and numerous inflammatory cells comprising mostly lymphocytes, plasma cells, few eosinophils. Univacuolated and multivacuolated lipoblasts with hyperchromatic nuclei were present. Some of the lipoblast showed a signet-ring appearance. Focal haemorrhagic and necrotic areas were noted. Histopathology picture was consistent with inflammatory atypical lipomatous tumor. Immunohistochemistry showed S-100 positivity in lipoblasts and Vimentin positivity in the stromal cells and was negative for Smooth muscle actin.

DISCUSSION:

ALT and well differentiated liposarcoma are synonyms describing lesions that are identical, morphologically and karyotypically, and in terms of biological potential.[2] However, the use of the term ALT is more appropriate for lesions occurring in the trunk and the somatic soft tissues, as wide surgical excision is possible at these sites and there is no recurrence. In contrast, tumors arising in the retroperitoneum and mediastinum may be referred to as “well

differentiated liposarcoma "because complete surgical excision is not possible and they usually show recurrence.

The main differential diagnosis are benign lipoma, and inflammatory myofibroblastic tumour. Lipomas are composed of mature adipocytes, which are uniformly arranged and show mimimal variation in cell size. In contrast to benign lipomas, adipocytes in ALT show significant variation in cell size. The adipocytes may show nuclear atypia and hyperchromasia and there may be presence of scattered hyperchromatic, multinucleated stromal cells. A characteristic hallmark of liposarcomas is the presence of univacuolated or multivacuolated lipoblasts, although WDL/ALTs do not always contain lipoblasts. At times the extensive lympho-plasmocytic infiltration mimics an inflammatory pseudotumour (inflammatory myofibroblastic tumour). Inflammatory myofibroblastic tumour do not have the atypical adipocytic component, which are found in atypical lipomatous tumour and they show cytoplasmic reactivity for ALK protein in 50-60% of cases. Also the spindle cells in inflammatory pseudotumour show variable positivity for SMA. Our case was negative for SMA. Inflammatory malignant fibrous histiocytoma (undifferentiated pleomorphic sarcoma) may also be considered in the differential diagnosis of inflammatory atypical lipomatous tumour. However, there is less cytological atypia in the spindle cells of the latter. Thorough sampling of the different regions of the tumour is essential to avoid misdiagnosis.

Most cases of inflammatory atypical lipomatous tumor have been reported in the retroperitoneum[3,4,5]. Two cases have been reported in the thigh[6]. Individual case reports have also been repored in the pleura,[7] scrotum[8], and the spermatic cord[9].

Atypical lipomatous tumour do not metastasize. However, tumor location greatly influences their rate of local recurrence and disease-related mortality. Tumors that occur in the extremities have much lower rates of local recurrence than those in the retroperitoneum. As complete excision is not possible for retroperitoneal tumors, they have a substantial risk of undergoing dedifferentiation (about 10-15%); which is somewhat lower for lesions in the extremity (5%)[10]. The overall mortality for atypical lipomatous tumor of the extremities may be 0%; while for well differentiated liposarcomas occurring in the retroperitoneum the mortality may be as high as to > 80%[1].

Figure 1: Gross specimen showing an encapsulated mass

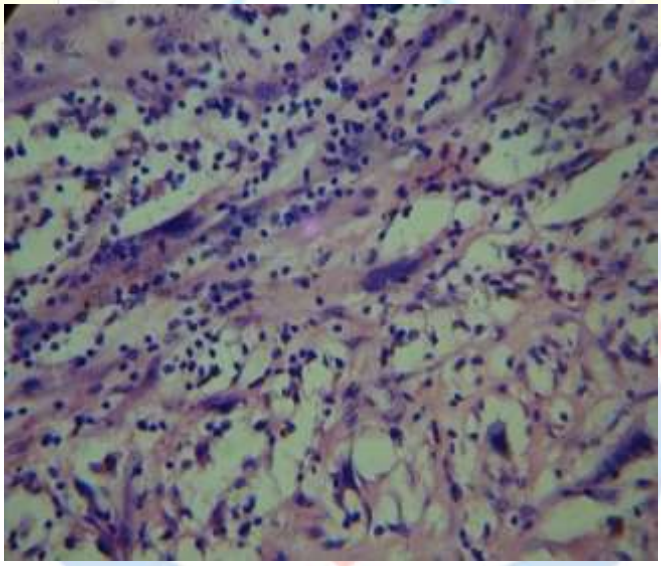

Figure 2: H & E stain showing lipoblasts admixed with inflammatory cells

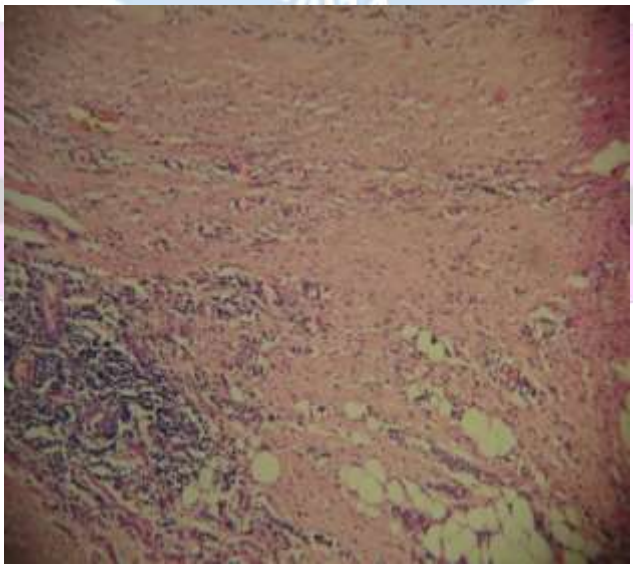

Figure 3: H & E stain showing adipocytes admixed with inflammatory cells

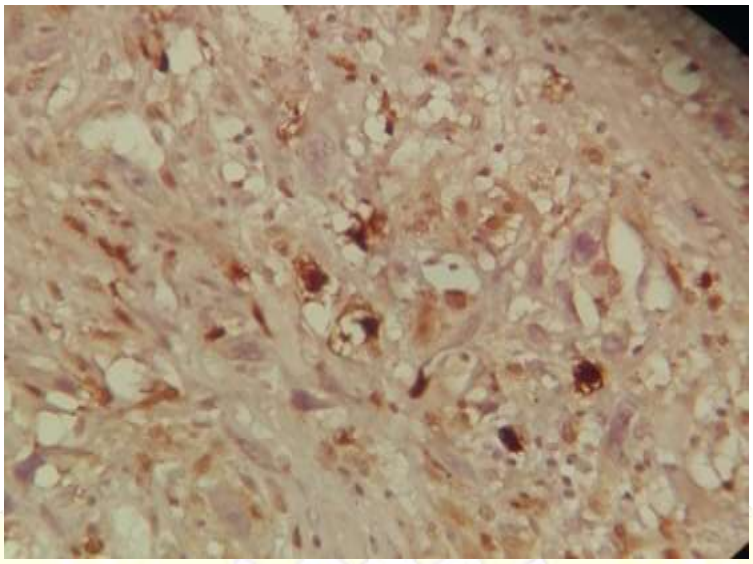

Figure 4: IHC showing S-100 positivity

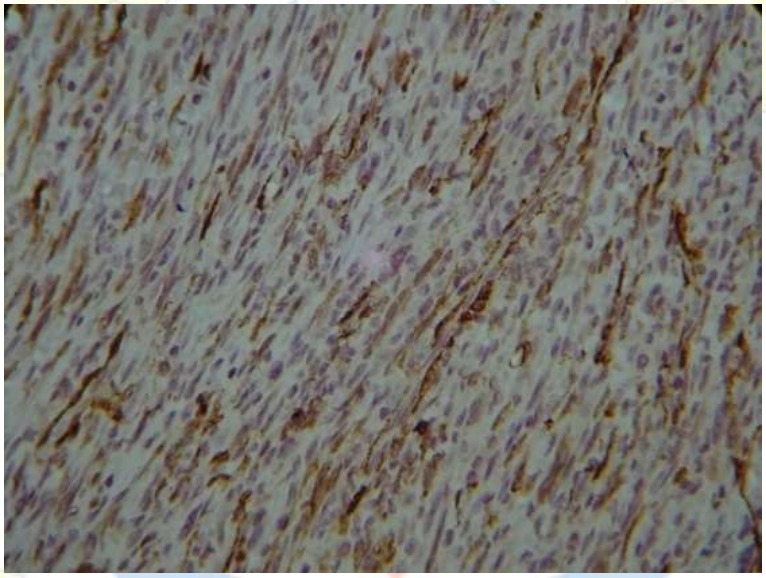

Figure 5: IHC, tumor cells showing vimentin positivity

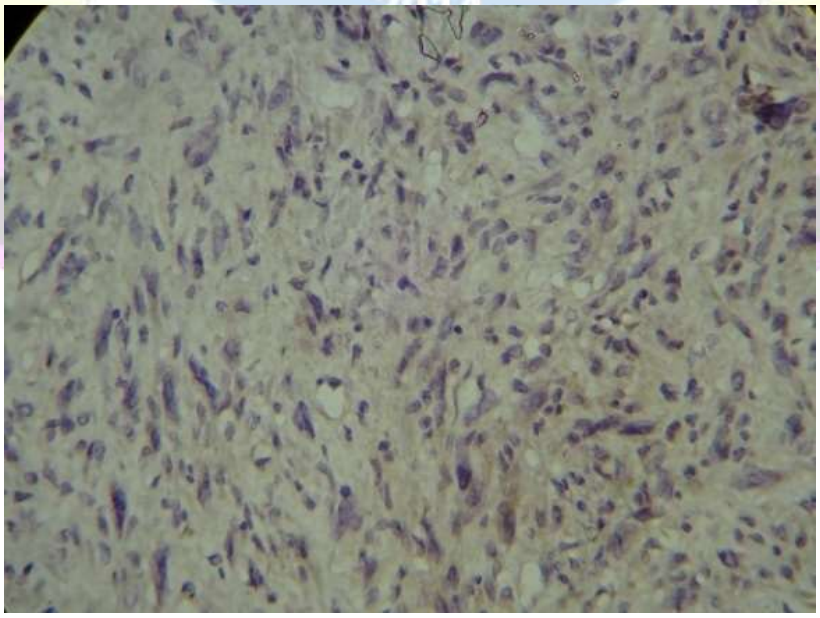

Figure 6: IHC, tumor showing absent staining for SMA.

CONCLUSION:

The primary treatment of all types of liposarcoma is surgical excision. The most important prognostic indicators are tumor location, size, and histologic subtype. In conclusion, inflammatory variant of well differentiated liposarcoma is a rare tumour found mainly in the lower limb and retroperitoneum. Awareness of this rare variant of well-differentiated liposarcoma is necessary for accurate diagnosis.

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