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Angiosarcoma of Anterior Abdominal Wall : A Rare Entity

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KEYWORDS

Vascular tumor, abdominal wall, abdominal lumps, histopathology, immunohistochemistry

ABSTRACT

Primary angiosarcoma is a rare malignant tumor originating from mesenchymal tissue, originating from endothelial cells and lymphatic vessels. In present case report, we reported a rare case of angiosarcoma at anterior abdominal wall in elderly male. On the basis of clinical findings and USG findings an elective excision biopsy was performed. Histopathological impression was poorly circumscribed high grade highly infiltrative malignant tumor with ramifying vascular pattern, compatible with malignant angiosarcoma tumor of vascular origin.Although rare, angiosarcoma should be included as differential diagnosis in cases of mobile abdominal lumps. Histopathology and immunohistochemistry are very important tools for diagnosis and should be considered in such cases.

1. Introduction

Primary angiosarcoma is a rare malignant tumor originating from mesenchymal tissue, originating from endothelial cells and lymphatic vessels. Its etiology is still unknown, but increasing evidence suggests the presence of characteristic molecular changes, which may be related to complex mechanistic pathway dysregulation leading to vascular growth disorders.[1] Angiosarcomas which account for only 1% to 2% of all soft tissue sarcomas are rare malignant tumors of endothelial origin.[2] These tumors have predilection for skin and superficial soft tissue, breast, bone, liver; they are rarely seen in deep soft tissue.[3] Predisposing factors for angiosarcoma include trauma, chronic lymphedema, irradiation, and age.[4,5] In present case report, we reported a rare case of angiosarcoma at anterior abdominal wall in elderly male.

2. Methods

A 74 years old, male patient was admitted in surgery department at SRM Medical College Hospital & Research Centre, Kattankulathur, Tamil Nadu, India. Patient came with swelling in the right anterior abdominal wall since 20 days, sudden in onset, Progressive in nature & associated with pain. No history of fever, trauma & no other complaint. No known Comorbidities. Bladder / Bowel habit normal. Normal sleep cycle. On examination, patient was conscious & oriented. His BP was 130/80 mm of hg, pulse rate – 86/min, cardiovascular system – S1, S2 normal, respiratory system – air entry bilaterally equal, per abdomen – soft, bowel sound present. A swelling of size 4 X 4 cm in the right hypochondrium, globular in shape, hard in consistency, tenderness present, irregular surface, well defined borders, mobile, leg raising & Head Raising positive (Swelling becomes Prominent). USG abdomen was done. A heterogeneously hypoechoic lesion measuring 2.4 x 1.1 cm noted in the deep subcutaneous / muscle plane in the anterior abdominal wall of the right side (region of interest). The lesion shows spindle configuration. Mild polar vascularity noted. Surrounding tissue shows inflammatory changes - possibility of intramuscular collection / abscess to be considered. On the basis of clinical findings and USG findings an

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elective exploratory Laparotomy was performed. The tumor along with portion of muscles was resected & resected specimen was sent for histopathology. Histopathological examination was done. Microscopic features showed fibrocollegerous and fibromuscular tissue showing an infiltrating neoplasm arranged in lobular pattern and composed of anastomosing vascular channels of varying caliber lined by atypical endothelial cells with pleomorphic vesicular nuclei and conspicuous nucleoli. 6-10 mitotic figures per high power field noted. Intervening areas shows extravasated RBCs, desmoplasia and hemorrhage. Adjoining skeletal muscle bundles are infiltrated by tumour cells. Histopathological impression was poorly circumscribed high grade highly infiltrative malignant tumor with ramifying vascular pattern, compatible with malignant angiosarcoma tumor of vascular origin, possibly angiosarcoma. Immunohistochemistry markers for confirmation were done.

FLI 1 - Positive in 90% of tumour cells.

Ki67 - Positive in 70-80%.

ERG - Positive in the atypical endothelial cells.

CD 31 - Positive in atypical endothelial cells

Histomorphology and IHC features are consistent with angiosarcoma

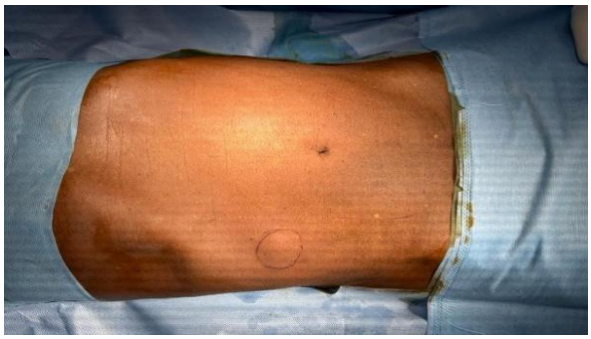

Fig 1 : Preop Picture of Anterior Abdominal wall lump

Fig 2 : Preop Picture of Anterior Abdominal wall lump

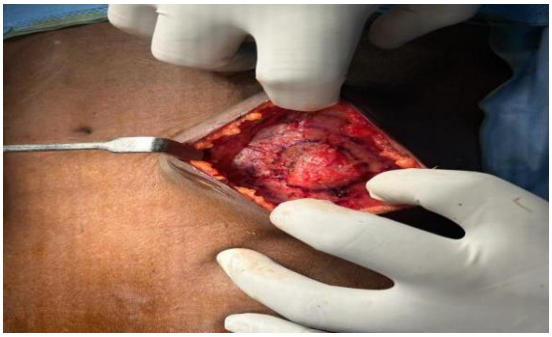

Fig 3: Intra op pictures

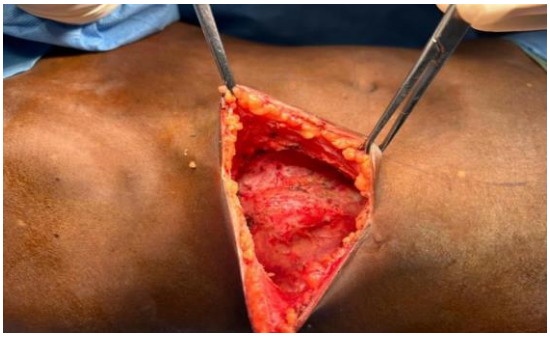

Fig 4: Intra op pictures

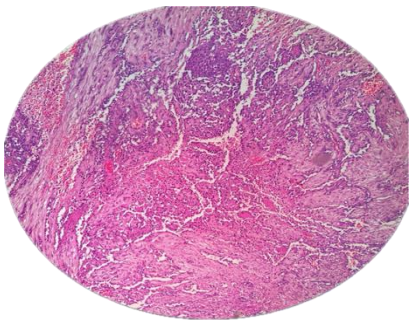

Fig 5 : Histopathology slide showing anastomosing vascular channels surrounding blood vessels

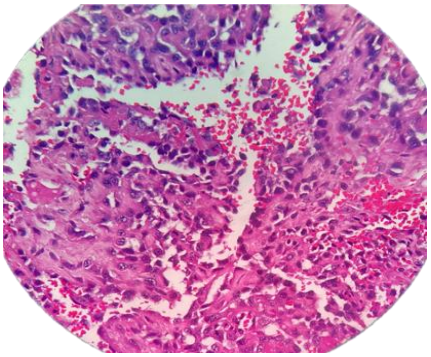

Fig 6: Histopathology slide showing tumor cells in high power field around blood vessels

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3.Discussion

Angiosarcoma is defined as the rapid and extensive infiltrative overgrowth of vascular endothelial cells. It resembles local invasion with high possibility of involvement of lymph nodes and metastasis. High expression of vascular specific receptor tyrosine kinases including TIE1, KDR, TEK, FLT, and VEGF leads to endothelial cell expansion, angiogenesis, and vascular leaks.

Due to its wide range of plausible etiologies, it has been categorized into lymphedema-associated, radiation-induced, primary breast angiosarcoma, sporadic cutaneous angiosarcoma (age related), commonly located on the head and neck region,[4] and angiosarcoma of the soft tissue.[5] Angiosarcomas are often misdiagnosed owing to their rarity and their subtle and varied clinical presentation. Pre-operative diagnosis of angiosarcoma by fine needle aspiration or core needle biopsy may be difficult due to insufficient sampling of the tumor.

The lower abdominal wall is an exceptional location for angiosarcoma,[7,8] and was only rarely described in case reports and small series.[9] In the largest prospective study, the Memorial Sloan-Kettering Cancer Center group identified 85 cases of soft tissue tumors of the abdominal wall from 1982 and 1999. No case of angiosarcoma was documented.[10]

Eric B et al.,[11] published a case report of very aggressive angiosarcoma of the abdominal wall in the context of repeated liraglutide subcutaneous injections, a glucagon-like peptid-1 receptor agonist, effective for weight management and glycemic control in type 2 diabetes, questioning a possible causal relationship between liraglutide and angiosarcoma.

Early detection by means of biopsy offers the only realistic chance of a cure.[12]Diagnosis is based on the microscopic features of the biopsy or specimen and the ultrastructural and histochemical markers. This aggressive malignant tumor of the vascular endothelium is characterized by rapid proliferation and infiltrating growth patterns.[13]

Morgan et al.,[12] has shown that the most common histologic pattern was characterized by anastomosing dissecting sinusoids lined by atypical endothelial cells (64%) with 15% of cases showing a diffuse epithelioid or spindle cell proliferation and 21% showing a mixture of the 2 histologic patterns.[12]

Immunohistochemical markers include von Willebrand factor, CD34, CD31, Ulex europaeus agglutinin 1, VEGF, and factor VIII antigen.[8] The most sensitive and specific marker for endothelial differentiation is CD31. In our case, besides, CD31+ cells and FLI1 cells, Ki67, and ERG was also positive.

Given its high risk of local recurrence and distant metastasis, the tumor portends a poor prognosis.[14] Treatment is usually by radical excision and reconstruction. Radiotherapy and chemotherapy are used for those with distant metastasis or unresectable tumors. Complete surgical resection of the tumor is the mainstay of treatment as there is a lack of consensus regarding systemic therapies.[15,16]

4.Conclusion

Although rare, angiosarcoma should be included as differential diagnosis in cases of mobile abdominal lumps. Histopathology and immunohistochemistry are very important tools for diagnosis and should be considered in such cases. As these tumors are very aggressive in nature, early and prompt diagnosis can help us in providing timely treatment in these patients.

Conflict of Interest: None to declare

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