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Research Paper Pathology

Inflammatory myofibroblastictumour of anterior abdominal wall in an adult.

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ABSTRACT

Inflammatory myofibroblastictumor (IMT) are rare proliferative, histologically distinctive lesion clinically resembling a malignant neoplasm with uncertain behavior. It is most commonly seen in the soft tissue of children and young adults, especially between 2–16 years. IMT rarely occurs in adults and the most commonly involved site is the lung, although there are a few case reports of extrapulmonary IMTs. According to our knowledge this is the second case of an adult with IMT of anterior abdominal wall. It is often misdiagnosed as a malignant lesion and frequently mismanaged. The diagnosis and treatment is a multidisciplinary approach. A thorough workup is necessary to avoid treatment-associated morbidity and mortality.

KEYWORDS : adult,anterior abdominal wall,Inflammatory myofibroblastictumour.

Introduction

Inflammatory myofibroblastic tumor (IMT) are rare proliferative, histologically distinctive lesion clinically resembling a malignant neoplasm with uncertain behavior.[1-4]It has also been previously known as inflammatory pseudotumor, plasma cell granuloma, inflammatory myofibroblastoma, inflammatory myofibrohistiocytic proliferation, mixed hamartoma, and inflammatory fibrosarcoma.[5,6]IMT frequently recurs and rarely metastasizes.[7,8]It is most commonly seen in the soft tissue of children and young adults, especially between 2–16 years.[6,8,9,10] IMT rarely occurs in adults, and the most commonly involved site is the lung, although there are a few case reports of extrapulmonary IMTs.[8,10,11]According to our knowledge this is the second case of an adult with IMT of anterior abdominal wall.

Case report

A 35-year-old male patient came with a mass over the anterior abdominal wall since 5 yrs gradually progressive in size.Clinical examination revealed a nontender, firm pedunculated mass about 10 cm in size. Clinical diagnosis of pedunculatedlipoma was made andsurgery was performed . Excised specimen was globular skin covered soft tissue mass measuring 11X7.5X4.2 cm. Cut surface was grey-white. (figure 1)Histopathology revealed cytologically bland spindle-shaped cells arranged in hyaline and myxoidstroma with an infiltration of lymphoplasmacytic cells.(figure 2)

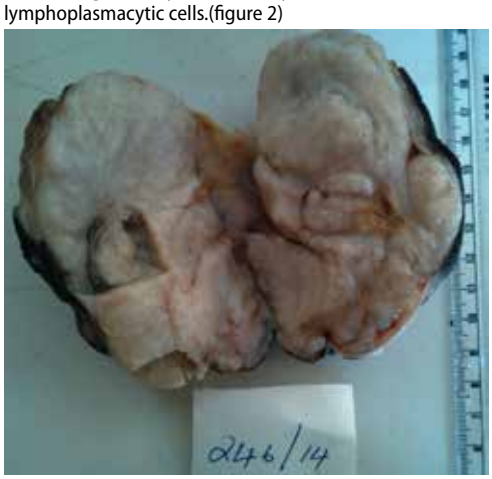

Figure 1. Photograph showing grey-white cutsurface.

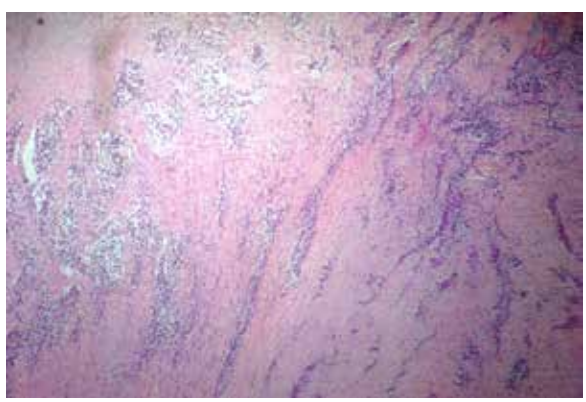

Figure 2. Photomicrograph showing spindle cells with inflammatory cells in hyalinised and myxoidstroma.

Discussion

Inflammatory myofibroblastic tumor is a rare spindle-cell lesion of intermediate malignant potential. [4]Inflammatory pseudotumor literally means an enlargement mimicking a tumor arising as a result of inflammatory accumulation of fibrous and granulation tissue as well as inflammatory cells.[12,13]It can occur in both pulmonary and extrapulmonary tissues.[4]IMTs can rarely involve the abdomen and most of the cases occur in the small bowel, colon mesentery, liver, spleen, retroperitoneum and othergastrointestinal sites.[6,12,14]

Pratap A et al reported paediatric IMT of anterior abdominal wall in 6-year-old boy.[11] Yagci MAreported the first case of an adult patient with anterior abdominal IMT presenting as an abdominal wall mass suggesting rhabdomyosarcoma.[5]To our knowledge, this is the second adult case of IMT of the abdominal wall. Females are affected slightly more commonly than males.[5,6]

The etiology of IMT is not clear, but the proposed mechanisms are prior allergic, immunologic, and infectious etiologies, and possibly foreign body reactions.[8,12,15] Some studies suggest a relation to infection and others a potential for low-grade neoplasia. [12]Although the etiology remains poorly understood, and it shows occasional local aggressiveness and the multifocality, thisentity uniformly follows a benign course.[12,16]

Clinical features depend upon the site and size of the lesion.[12] Clinical symptoms of abdominal IMT are not specific to the disease and depend on localization and growth pattern of the tumor and is usuallypalpable mass, abdominal pain, weight loss or fever.[6,15]The other laboratory findings are often inconclusive which include microcytic hypochromic anemia, leukocytosis, thrombocytosis, hypergammaglobulinemia, and high sedimentation rate.[6,9]Computed tomography is often suggestive of a tumor.[12]

In most cases, a definitive diagnosis is made based on the histopathology. IMT is Typically, it is a circumscribed but nonencapsulated lesion microscopically composed of myofibroblasticspindle cells, with an inflammatory cell infiltrate of plasma cells, lymphocytes and eosinophils, and different patterns can be found within the same tumor.[4,6,8,10,15] The histopathological subtypes arefibromyxoid and vascular pattern, proliferating pattern, and sclerosing pattern.[5,6] In IMTs, high cellularity with large, plump, active myofibroblasts withprominent nucleoli can cause confusion with malignancy, in particularrhabdomyosarcoma. However the lack of atypia, hyperchromasia and abnormalmitotic figures are pointers toward benign lesion.[4] Immuhistochemistry is adjunct to diagnosis which confirms the predominant cell as myofibroblast[6]. Vimentin is almost invariably positive in the spindle cells. Smooth muscle actin, muscle-specific actin and desmin are present in majority of the cases. CD68 (KP-1), CD30 (Ki-1), cytokeratin and p53 are positive in some cases.[6]. Su et al showed some monoclonal chromosomal anomaly in three cases that support the neoplastic nature of this lesion.[6,17,18]Surgical excision is the only treatment for IMT [6]Complete excision is necessary to avoid local recurrence. Local recurrences after incomplete excision are recognised, may occurmany years later and may be fatal. This underlines the importance of completesurgical resection whenever possible.[4]

Conclusion

In conclusion, inflammatory myofibroblastic tumors of the abdominal wall are rare entities in adults. It is a benign disease that is often misdiagnosed and frequently mismanaged.The diagnosis and treatment is a multidisciplinary approach. A thorough workup is necessary to avoid treatment-associated morbidity and mortality2

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