Case Report

Inflammatory myofibroblastic tumor of the liver: A rare pathological entity

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ABSTRACT

Inflammatory myofibroblastic tumor (IMT) of the liver is a rare tumor like lesion that can mimic malignant liver neoplasm. IMT is a lesion composed of myofibroblastic spindle cells, plasma cells, lymphocytes, and eosinophils. It can occur in soft tissues and viscera. It was previously called plasma cell granuloma, inflammatory myofibrohistiocytic proliferation, and inflammatory pseudo tumor, but IMT is the designation currently used. IMT is more frequently described in the lung and abdomen of young patients, but it can also be found in the central nervous system, salivary glands, larynx, bladder, breast, spleen, skin, and liver. Here, we present a case of IMT of the liver in a 55-year-old female patient which is a rare presentation.

Key words: Hepatectomy, inflammatory myofibroblastic tumor, liver

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) of the liver is an uncommon lesion of uncertain pathogenesis that has a unique histological appearance. The symptoms and radiological findings of this rare tumor can pose diagnostic difficulties. Pack and Backer published the first case occurring in the liver.[1] Since then, reports on IMT in the liver with different progression have been described.[2] Concerning pathogenesis, it is controversial whether IMT is a neoplasm or a reactive pseudotumoral lesion.[3] The main aim of presenting this case is to highlight this rare tumor and also to consider IMT as a differential diagnosis clinically and radiologically for any liver neoplasm before concluding it as a malignancy.

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CASE REPORT

A 55-year-old female patient came with chief complaints of pain in the abdomen more on the right side, fever, loss of weight, and jaundice since three months. There was no abdominal lymphadenopathy and no ascites. Liver function tests showed raised levels of total bilirubin; other parameters were almost normal. Radiologically and clinically, it was diagnosed as liver metastases from an unknown primary. There were raised levels of erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP). Explorative laparotomy with extended left hepatectomy was done and sent for biopsy. Grossly, we received a hepatectomy specimen of weight 575 g, size 10×6×4cm, firm to hard, and externally gray-brown. Cut section showed a large variegated lesion, well circumscribed and multinodular measuring 7×5cm [Figure 1]. Multiple sections were taken from the lesion and it was confirmed as IMT of liver by immunohistochemistry (IHC).

DISCUSSION

IMT is a benign, tumor like mass characterized by proliferating fibrous tissue infiltrated by inflammatory cells.[4] This condition is known to occur in several organs including lymph nodes, spleen, brain, spinal cord, larynx, thyroid gland, breast, pancreas, gastrointestinal tract, and bladder, but most frequently it occurs in the lungs and liver.[5] Hepatic IMTs occur predominantly in the right lobe of the liver, although multicentricity has been described,[6] and it has previously occurred in the gall bladder fossa of the liver.[7]

Laboratory investigations often indicate an ongoing inflammatory process with a leukocytosis and a raised ESR and CRP, as was evident in the present case. In the present case, the levels of total bilirubin were raised, with other parameters in normal range.

Microscopically, the lesion showed a mass of fibroblastic and myofibroblastic cells set in a loose myxoid matrix

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containing scattered lymphocytes, plasma cells, eosinophils, and histiocytes consistent with a myofibroblastic tumor [Figure 2].

Smooth muscle actin (SMA) and vimentin are usually positive in stellate and spindle cells, whereas desmin, CD34, S100 protein, and anaplastic lymphoma kinase are negative.[8,9]

In the present case, IHC was positive for leukocyte common antigen (LCA), SMA [Figure 3a and b], and vimentin [Figure 4], and it was finally confirmed as Myo Fibroblastic Tumour (MFT) of liver.

It is likely that many spindle cells correspond to activate histiocytes as they co express vimentin and macrophage-associated markers; they are intermingled with vimentin-positive fibroblasts and variable numbers of vimentin- and actin-positive myofibroblasts. Because of the variable immunophenotypic patterns seen in hepatic IMT, it is possible that they arise from a common mesenchymal cell that is capable of differentiating along different pathways. The majority would develop a myofibroblastic phenotype and be positive for SMA and vimentin. Many SMA-positive myofibroblastic cells were found in IMT, hence suggesting an ongoing fibrous process.

The management of an IMT of the liver has traditionally been surgical, but more recently spontaneous regression of the tumor has been reported with the use of antibiotics or nonsteroidal anti-inflammatory drugs.

IMT of the liver is a rare benign neoplasm and is often mistaken for a malignant entity. Few cases have been reported in the literature and the precise etiology of the inflammatory pseudotumor remains unknown. Patients usually present with fever, abdominal pain, and jaundice. The proliferation of spindled myofibroblast cells mixed with variable amounts of reactive inflammatory cells is characteristics of IMT. IMT of the liver constitutes a diagnostic and therapeutic challenge. In the presence of a solitary liver lesion, with clinical and laboratory features suggesting active inflammation, the diagnosis of inflammatory pseudo tumor should be considered. A proper investigation to exclude malignancy should be undertaken, and resection of the lesion should be considered when in doubt.

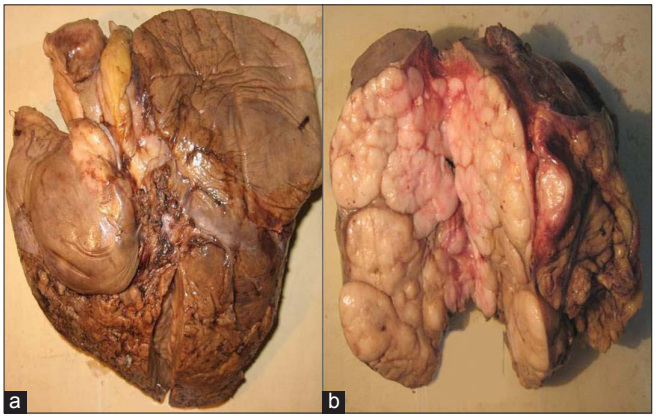

Figure 1: (a) Gross showing hepatectomy specimen; (b) cut section showing a large variegated lesion, well circumscribed and multinodular lesion measuring 7×5cm

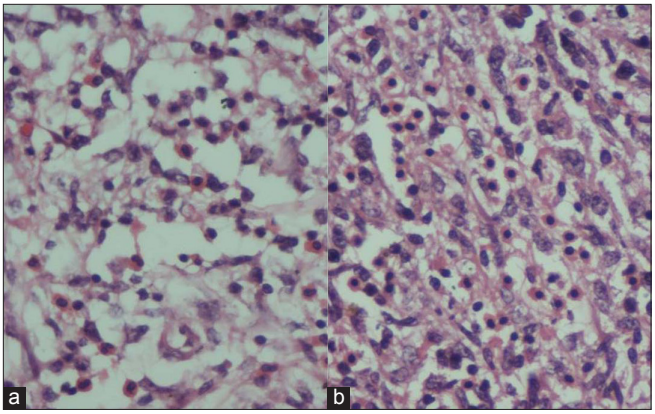

Figure 2: (a and b) Sections showing fibroblastic and myofibroblastic cells set in a loose myxoid matrix containing scattered lymphocytes, plasma cells, eosinophils, and histiocytes (H and E, ×10)

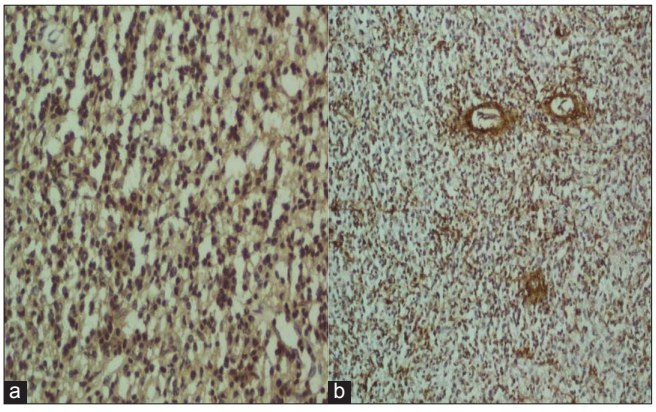

Figure 3: (a) Immunohistochemistry showing positivity for leukocyte common antigen and (b) smooth muscle actin

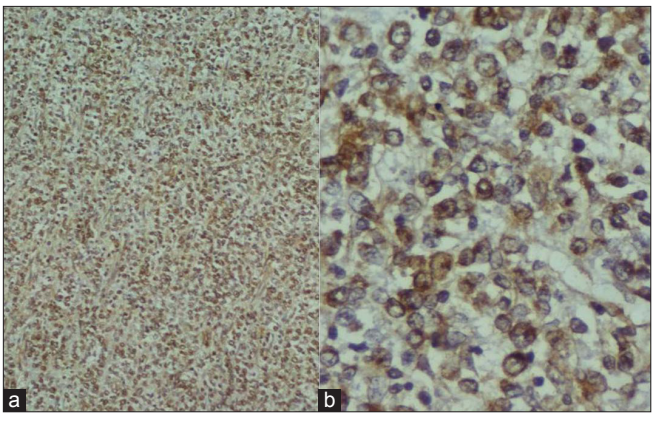

Figure 4: (a and b) Immunohistochemistry showing positivity for vimentin

In our case, the lack of atypical mitoses and cellular atypia together with histopathological and immunohistochemical findings supported the diagnosis of IMT.

In conclusion, this case report emphasizes the importance of considering IMT as part of the differential diagnosis, and highlights that a conservative approach to a patient with IMT is an effective treatment option.

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