Inflammatory myofibroblastic tumor of the gall bladder: a rare case report

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Keywords: Inflammatory, Myofibroblastic Tumor, Gall Bladder, Pseudotumor.

Abstract

Inflammatory myofibroblastic tumor (IMT) is a benign proliferation of myofibroblasts, which is prone to recurrence, persistent local growth as well as malignant transformation. The commonest site reported in the biliary tree is the extrahepatic bile duct. Gall bladder is a rare site for the development of an IMT. We report a case of an inflammatory myofibroblastic tumor of the gall bladder in an adult female with coextistent cholecystitis and cholelithiasis. It is important to acknowledge this entity since it can mimic malignancy.

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Date of Submission: June 29, 2014       Date of Acceptance: July 18, 2014       Date of Publishing: Jan 28, 2015

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Introduction

Inflammatory myofibroblastic tumor (IMT), also known as inflammatory pseudotumor or plasma cell granuloma, is a rare benign lesion which has been described in various sites of the body including intrabdominal locations. The age range is broad, but IMT has a predilection for children. Most common intrabdominal location is the mesentry and the omentum. It has been frequently reported in the liver, stomach, intestine, spleen, mesentry and biliary tree.\(^\text{1,2}\) Gall bladder is a rare site for the development of an IMT. We report a case of an IMT of the gall bladder in an adult female.

Case Report

A 42-year-old female presented to the surgery OPD with off and on colicky pain in the right upper quadrant of the abdomen for the past 6 months. The pain was sudden in onset and referred to the tip of the right shoulder. Routine investigations were planned out. Complete blood count was normal except for haemoglobin which was 10.6g/dl. On ultrasound, multiple stones were found in the gall bladder and the wall was diffusely thickened at the fundus. Open cholecystectomy was carried out 1 week later. Per operatively, gall bladder was adherent to the omentum, had a thickened wall with pus and multiple stones in the lumen.

On gross pathological examination, external surface of the gall bladder was irregular, shaggy and congested. The fundus of the gall bladder was adherent to fibrofatty tissue (? omentum). On cutting open, multiple mixed stones were found in the lumen. Mucosa was ulcerated and focally congested. Wall thickness varied from 0.3 cm in the body to 1cm at the fundus.

Microscopically, the sections showed mucosa, fibromuscular layer and adventitia of the gall bladder. Mucosa was ulcerated. The fibromuscular layer and adventitia were focally replaced by proliferative spindle shaped myofibroblastic cells which were arranged in intersecting fascicles in a haphazard manner. (Figure 1, 2) These cells were plump, ovoid to spindle shaped with eosinophilic cytoplasm, round to ovoid nuclei with vesicular chromatin and a prominent nucleolus. Mitotic figures were inconspicuous. There was marked infiltration by lymphocytes, plasma cells and histiocytes in this area. (Figure 3) Focally lymphoid aggregates were present. The lesion involved the surrounding fat and liver bed.

Immunohistochemistry was performed and the spindle shaped cells were positive with vimentin and SMA (Figure 4), faintly positive with desmin, negative for
S-100 and chromogranin. CD 68 was positive (Figure 5) in the histiocytic cells.

In view of the above findings, a diagnosis of IMT of the gall bladder was made. On follow up, the post operative course was uneventful. Computed Tomography of the chest and abdomen did not reveal any residual lesion or metastasis. The patient is currently asymptomatic.

Discussion
Inflammatory myofibroblastic tumor is a benign proliferation of myofibroblasts, which is prone to recurrence and persistent local growth. Intra-abdominal forms are most frequent in the liver [1] and the commonest site reported in the biliary tree is the extrahepatic bile duct. [2]

IMT is composed of fascicles of bland myofibroblastic cells admixed with a prominent inflammatory infiltrate consisting of lymphocytes, plasma cells, macrophages and eosinophils. The lack of atypia, hyperchromasia and abnormal mitotic figures are pointers towards a benign lesion. Histological picture varies from predominantly loosely arranged spindle-stellate cells in a myxoid or hyaline matrix with scattered inflammatory cells to a compact proliferation of cells in a storiform or fascicular pattern. The spindle cells stain positively for smooth muscle actin, vimentin and are negative for S100, desmin, CD10, CK, CD35 and latent membrane protein.

Such lesions fall into 4 main categories- reactive lesions, benign lesions, locally aggressive lesions and malignant lesions. [3] The favored pathogenesis for an IMT is an exaggerated response to tissue injury rather than an immunological phenomenon. [3] However, the exact etiology is unclear. [4,5] The term inflammatory pseudotumor has been replaced by inflammatory myofibroblastic tumor since the cell of origin was established to be a myofibroblast by immunohistochemistry and electron microscopy. [6]

Behranwala et al [3] and Muduly et al [7] reported 2 separate cases of IMT of the gall bladder which were clinically suspected to be an infiltrating gall bladder carcinoma. Another case of chronic cholecystitis with IMT was described by Corsi et al. [8] This case showed a xanthogranulomatous inflammation along with huge fibro and myofibroblastic proliferation. A case of IMT of the gall bladder with a synchronous lesion in the lung has been reported previously. [6] Ikeda et al reported an IMT of the gall bladder and bile duct in a patient with obstructive jaundice. [9] Gall bladder is a very rare site for the development of an IMT. The present case was one with coexistent cholecystitis with an inflammatory myofibroblastic tumor which was involving the surrounding omentum.

Conclusion
Differentiation of IMT from a malignant lesion is important in order to obtain a definitive diagnosis as well as to relieve symptoms. Management of such tumors is surgical for definitive diagnosis and for symptomatic improvement. Since local recurrence and malignant transformation has been reported, strict follow up is mandatory.

Acknowledgements
None

Funding
None.
Competing Interests
None declared.

References