



Inflammatory myofibroblastic tumor involving liver, gallbladder, pylorus & duodenum: A rare case presentation

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ABSTRACT

Inflammatory myofibroblastic tumors are rare benign tumors that can mimic malignancy of unknown aetiology. It has spectrum of myofibroblastic proliferation along with varying amount of inflammatory infiltrate. Recently, the concept of this lesion being reactive has been challenged based on the clinical demonstration of recurrences and metastasis and cytogenetic evidence of acquired clonal chromosomal abnormalities. We hereby report a case of inflammatory myofibroblastic tumor involving liver, gallbladder pylorus and 1st part of duodenum.

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1. Introduction

Inflammatory myofibroblastic tumor (IMT) occurs more frequently in childhood and the most common involvement is seen in the lungs. Different terms have been applied to the lesion, namely, inflammatory pseudotumor, fibrous xanthoma, plasma cell granuloma, pseudosarcoma, lymphoid hamartoma, myxoid hamartoma, inflammatory myofibrohistiocytic proliferation, benign myofibroblastoma, and most recently, inflammatory myofibroblastic tumor [1]. Primary inflammatory myofibroblastic tumors of the gallbladder are rather infrequent. The present knowledge is based on case reports.

IMT was first observed in lungs and described by Bunn in 1939. It was named as IMT by Umiker et al. because it mimics malignant neoplasm clinically, radiologically and histopathologically [1]. Initiating factors such as reactive infections, autoimmune and neo-

plastic processes, has been proposed but the aetiology of most remains unknown.

2. Presentation of case

We present a case of 36 yrs old female who came to surgery OPD having chief complaint of bloating of abdomen and dyspepsia for 5 months. On examination her vitals were stable. On abdominal examination it was Soft, no organomegaly noted. Routine Blood investigation were within normal limits. Ultrasonography of whole abdomen revealed gall bladder calculi with asymmetric wall thickening (9 mm). The interface with liver is hazy. As noted by ultrasonography of having asymmetric wall thickening CT SCAN was done. CT SCAN abdomen report revealed-GB shows mild diffuse wall thickening with asymmetric wall thickening in body region extending along the adjacent duodenum with circumferential thickening of the duodenal wall causing slight luminal narrowing with mild proximal gastric distension. Fat plane between GB & lesion poorly visualised with mild hepatic altered attenuation (depth upto 1 cm). No significant biliary dilation (Fig. 1 (A,B,C)). As the patient was only presenting with bloating of abdomen & dyspepsia, without any significant features of obstruction, upper GI endoscopy was done to rule out any hiatal disorders, ulcers &

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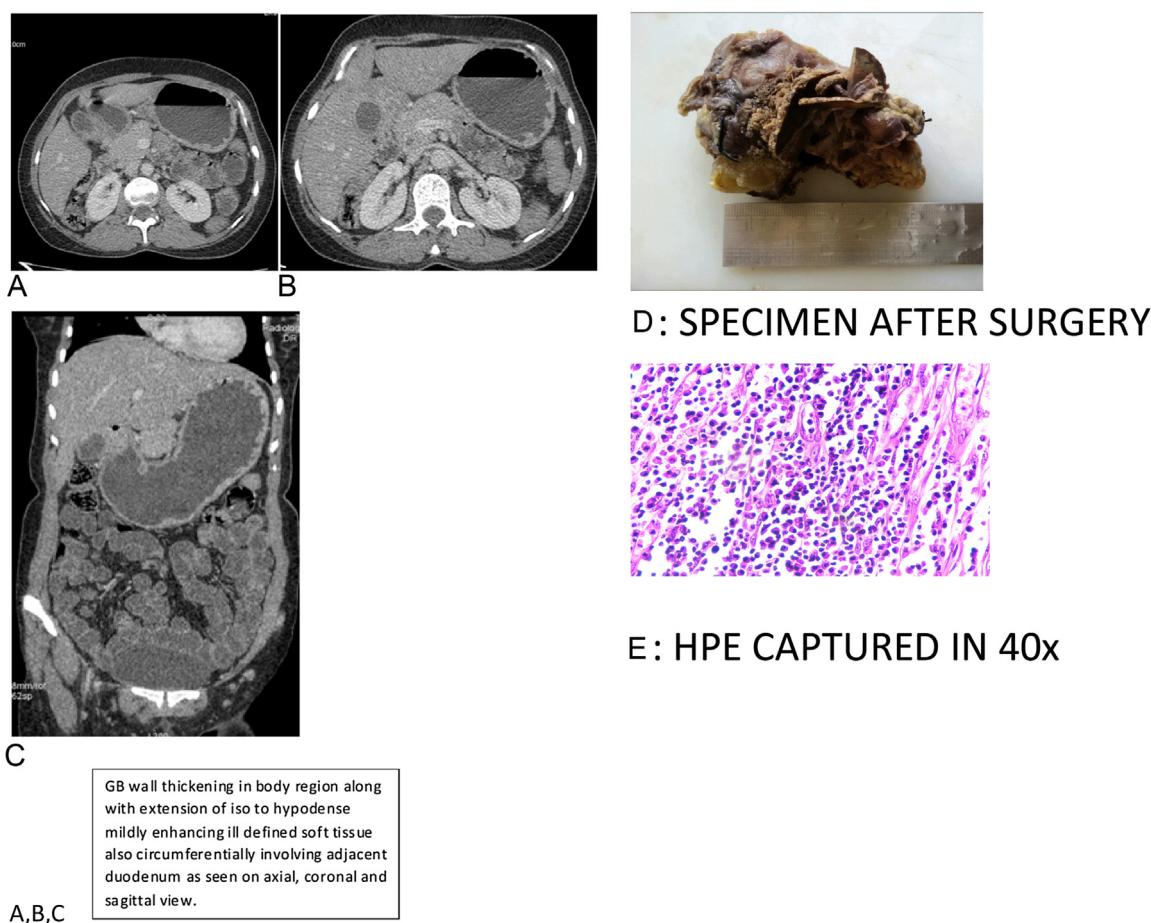


Fig. 1. (A,B,C) GB wall thickening in body region along with extension of iso to hypodense mildly enhancing ill defined soft tissue also circumferentially involving adjacent duodenum as seen on axial, coronal and sagittal view. (CT SCAN).

to take biopsies if possible. Upper GI endoscopy was also normal. Hence planing was to do Extended cholecystectomy. But intraoperatively it was found that there were lots of adhesion at gall bladder-liver interface & pylorus on palpating gallbladder wall appeared thick with full of calculi. There was thick & hard mass like feeling at pylorus which was extending to 1st part of duodenem. Hence, Extended cholecystectomy with resection of pylorus and 1st part of duodenum with peripancreatic lymph node dissection, billrothII anastomosis done under general anaesthesia & specimen sent for HPE (Fig. 1 D). Histopathology shows-Inflammatory myofibroblastic tumor with involvement of liver & resected end of pylorus & duodenum with associated chronic cholecystitis (Fig. 1 E).

Contribution of all athours were there, as from preoperative preparation to postoperative outcome, as the patient was discharged from hospital unevenfully after histopathology report, & instructed to follow up.

3. Discussion

Inflammatory myofibroblastic tumors are rare benign structures with unknown aetiology. IMT occurs mainly in children and young adult. IMT in the pediatric abdomen have clinical importance because the lesion often mimic malignant neoplasm, such as sarcoma, lymphomas, or metastases [2].

Lymphocytes, plasma cells, histiocytes, fibroblasts and myofibroblasts are the basic components of IMT, present in variable proportions.

Four basic histologic patterns are commonly seen as follows:

- a.) Dominant lymphoplasmacytic infiltrate;
- b.) Dominant lymphohistiocytic infiltrate;
- c.) Young and active myofibroblastic process and
- d.) Predominantly collagenized process with lymphocytic infiltrate.

The lymphoplasmacytic IMT consists of a mature lymphoid and plasma cell infiltrate with germinal centers, hence the name given is plasma cell granuloma [3]. Lymphohistiocytic IMT most commonly resembles an infectious process as foamy histiocytes are predominant [3]. Collagenized IMT is less cellular and resembles a desmoid tumor but with a prominent inflammatory infiltrate. A zonation/maturation effect may be observed. The most specific feature of IMTs, whose main cell type is myofibroblasts, is the ability to mimic malignancy. A histopathological examination of the IMTs revealed numerous lymphocytes, plasma cells and histiocytes settled between the spindle cells and fibrous tissues [4]. Although IMT was first described by Brunn, its pathological entities were identified in detail by Umiker and Iversen in 1954 [5]. Two theories have been emphasised to explain the aetiopathogenesis: the first is an abnormal host response to tissue damage, while the second is disorders in immunological responses [6,7]. IMTs are known to be benign tumors with no capacity to metastasise. However, these tumors are known to infiltrate surrounding tissues due to their severe proliferative capacity and they are thus known to recur frequently, even after resection [8].

The most frequently involved organ in the abdomen is the liver, while primary gallbladder involvement is quite rare. Present

knowledge about this entity is based on case reports in the literature [9]. The clinical picture in cases with IMT depends on the organ of involvement and site of the organ; abdominal pain, jaundice and ascites might be the presenting symptoms, according to the localisation in the liver involvement.

Gallbladder involvement, on the other hand, may result in acute cholecystitis, biliary colic, obstructive jaundice and cholangitis may be seen in cases with extension to the common bile duct. In our case symptoms were abdominal pain and dyspepsia. CT demonstrated involvement of liver, gallbladder, pylorus, & 1st part of duodenum, which is rare. Hence, intraoperatively Extended cholecystectomy + BillrothII gastrectomy done.

4. Conclusion

Inflammatory myofibroblastic tumor of liver, gallbladder, pylorus, duodenum is rare. Hence, surgery is the main treatment.

Conflicts of interest

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Consent

Yes.

Author contribution

Dr Lakshmi sinha – study concept, design & writing paper.

Dr Arsalan Hasan – writing paper.

Dr Akhilesh Kumar Singh – writing paper.

Dr Poonam Prasad Bhadani – pathology updates.

Dr Achuta Nand Jha – radiology interpretation.
Dr Prashant Kumar Singh – data collection.
Dr Manoj Kumar – data interpretation.

Guarantor

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