**Rare presentation of self-resolving multifocal inflammatory pseudo-tumour of liver**

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**Abstract**

Inflammatory pseudo-tumour (IPT) of the liver is a rare condition with the appearance of a tumour-like space occupying lesion. Aetiology and natural history is not known for these benign lesions, as they are commonly diagnosed as malignant lesions and frequently undergo surgical resection since spontaneous resolution is very rare. Multifocal IPT involving both lobes of liver are rarely reported. Here we report a unique case of multifocal IPT of the liver which resolved spontaneously within 5 wk period.

**Core tip:** Inflammatory pseudo-tumour (IPT) is extremely rare pre-operative diagnosis. It's a benign condition which does not require surgical management and it rarely self resolves. Our case so unique in appearance that IPT generally are solitary and if multiple generally confined on lobe of liver. In our case this tumour mimicked metastasis and presented in bilobar presentation. It resolved in 5 wk something which is never reported in medical literature.

**INTRODUCTION**

Inflammatory pseudo-tumours (IPTs) represent a challenging clinico-pathologic entity due to their similarity with malignant lesions. Histopathologically these tumours are inflammatory myofibroblastic tumours or plasma cell granulomas. They were initially observed in the lung by Brunn et al in 1939, and subsequently in the liver by Pack et al in 1953. Although in the liver they may rarely lead to biliary obstruction, portal hypertension and even cirrhosis, their clinical importance is primarily related to the difficult differential diagnosis from malignant tumours [1]. For this reason the main pattern of diagnosis is on post-resection histopathological processing. However, spontaneous resolution of these lesions has been reported as well. Here we present a case of multifocal liver IPT, which spontaneously resolved without any type of therapeutic intervention.
CASE REPORT

A 37-year-old Caucasian female was referred to our tertiary hepatopancreatobiliary surgical unit with symptoms of weight loss and malaise since one month. There are no pre-existing medical co-morbidities. Bloods results showed derangement of liver function tests with alkaline phosphatase of 174 U/L (normal range: 38-126), aspartate transaminase of 40 U/L (9-36), alanine transaminase of 43 U/L (10-28), and normal bilirubin. Regarding inflammatory markers only erythrocyte sedimentation rate was raised at 58 mm/h (2-12). Abdominal ultrasound revealed multiple bilobar lesions in the liver with maximum size of 3.5 cm adjacent to the porta hepatis. Multidetector computed tomography with a liver protocol demonstrated 5 lesions, non-enhancing and of low attenuation highly suspicious of metastatic origin (Figure 1). According to decision of the multidisciplinary team meeting, the patient was considered harbouring metastatic diseases of unknown primary and she underwent detailed investigations with upper gastrointestinal endoscopy, positron emission tomography (PET) scan, mammography, chest computed tomography scan and tumour markers as well. Apart from an abnormal uptake in the right lobe of the liver on PET scan there was no pathology revealed in any other study. After failure in locating the primary, a percutaneous biopsy of a liver lesion was decided as next step in the diagnostic workup. Histopathology revealed a parenchyma of normal architecture but replaced by collagenous stroma admixed with inflammation. The stroma was composed of dense areas of fibrosis, myofibroblasts and fibroblasts. The inflammation consisted of a mixture of plasma cells, lymphocytes and eosinophils. Aggregates of xanthomatous histiocytes were also seen. There was mild portal inflammation. Also few epithelioid granulomas were seen. Immunohistochemistry showed positivity of the spindle cells for smooth muscle actin and negativity for ALK-1 (Figures 2 and 3). Epstein barr virus and cytomegalovirus immunostains were negative as well. Ziel-Neelsen showed no evidence of acid-fast bacilli in specimen. The tissue sample was considered as highly suggestive of inflammatory pseudo-tumour.

Based on the existed evidence and according to patient’s consent, a wait and watch policy with follow-up imaging in 3 wk was decided. Magnetic resonance imaging (MRI) demonstrated significant reduction in the size of all liver lesions, measuring a maximum diameter of 1.5 cm as compared to the previous 3.5 cm. Follow-up MRI at two months showed almost complete resolution of the lesions. She was put on annual follow up with liver MRI and she remains asymptomatic and without evidence of any lesions on 2 year follow-up (Figure 4).

DISCUSSION

An IPT is a benign, tumour like lesion characterised by...
of tumour markers as CA19-9 might confuse further the final diagnosis, but this was not observed in our case.

In conclusion, IPT might represent a considerable diagnostic challenge requiring high index of suspicion. The characteristic of spontaneous resolution highlights the necessity of regular, short-term, imaging follow-up in order to avoid aggressive unnecessary interventions.

**COMMENTS**

**Case characteristics**
A 37-years-old female without any medical co morbidities presented with short term history of weight loss and malaise and diagnosed with multiple liver lesions.

**Clinical diagnosis**
Multiple liver lesions.

**Differential diagnosis**
Metastatic disease in liver, multifocal hepatocellular carcinoma, inflammatory pseudotumour.

**Laboratory diagnosis**
Bloods results showed derangement of liver function tests with alkaline phosphatase of 174 U/L (normal range: 38-126), aspartate transaminase of 40 U/L (9-38), alanine transaminase of 43 U/L (10-28), and normal Bilirubin. Erythrocyte sedimentation rate was raised at 58 mm/h (2-12). Other blood tests including tumour were normal.

**Imaging diagnosis**
Multi detector computed tomography with a liver protocol demonstrated 5 lesions, non-enhancing and of low attenuation highly suspicious of metastatic origin.

**Pathological diagnosis**
Histopathology revealed a parenchyma of normal architecture but replaced by collagenous stroma admixed with inflammation. The stroma was composed of dense areas of fibrosis, myofibroblasts and fibroblasts. Immunohistochemistry showed positivity of the spindle cells for smooth muscle actin.

**Treatment**
This patient did not require any treatment as inflammatory pseudo-tumour self-resolved completely and patient was completely asymptomatic even at 2 years follow up.

**Related reports**
There are very case reports of this particular condition but these lesions are more frequently diagnosed because of routine use of cross sectional imaging. It is important to be aware of this particular condition to spare patient of major surgical procedure.

**Experiences and lessons**
The biology pathophysiology and disease history of this particular condition is not completely known. This case report highlights importance of serial radiological examinations and unique place of fine-needle aspiration cytology in diagnosis of inflammatory pseudo tumours.

**Peer review**
This is a brief, easy to read and interesting case report in which the authors show a patient with a usual clinical presentation.

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