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Case Report

Extramedullary hematopoiesis (EMH) in the liver allograft presenting with a mass-like lesion ☆,☆☆

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ABSTRACT

This is a rare case of extramedullary hematopoiesis (EMH) presenting as a mass-like lesion in liver allograft. Our patient was a 57-year-old woman who had undergone liver transplantation due to hepatic epithelioid hemangioendothelioma. She presented with an ill-defined hypoechoic lesion on ultrasound which showed features of focal EMH on pathologic examinations. While transient intrahepatic hematopoiesis has been reported in liver transplant patients, focal EMH mass lesion is a rarely encountered phenomenon. Therefore, focal EMH may need to be considered as a differential diagnosis when encountering a mass in post liver transplant patients.

Introduction

Blood cells arise from hematopoietic stem cells which reside primarily in the bone marrow. This process is called hematopoiesis. Extramedullary hematopoiesis (EMH) is defined as the occurrence of hematopoiesis in organs other than bone marrow. Liver, spleen, and lymph nodes are the most common sites for EMH [1].

Here, we report a case of EMH in the liver allograft presenting with a mass-like lesion.

Case presentation

Our patient was a 57-year-old woman who had undergone liver transplantation due to hepatic epithelioid hemangioen-

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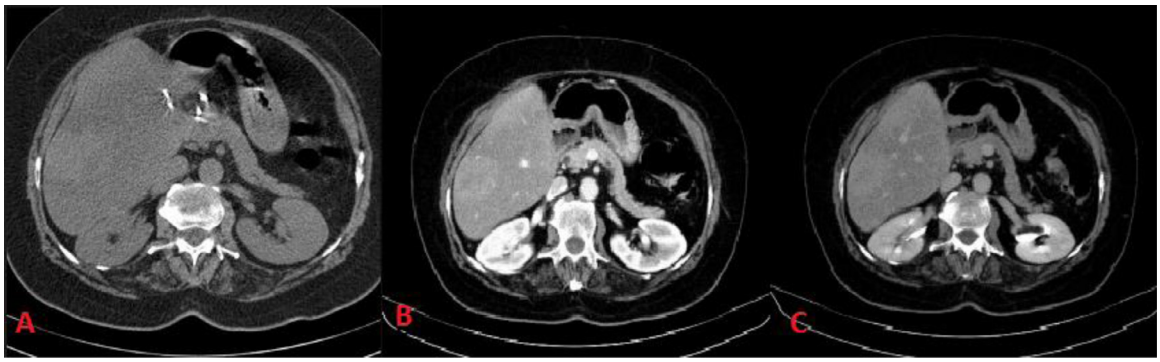

Fig. 1 – Hepatic CT shows an ill-defined lesion which is hyperdense to surrounding low-density fatty liver on noncontrast images (A). Contrast enhanced images show enhancement in arterioportal phase (B) with preserved enhancement in delayed phase (C).

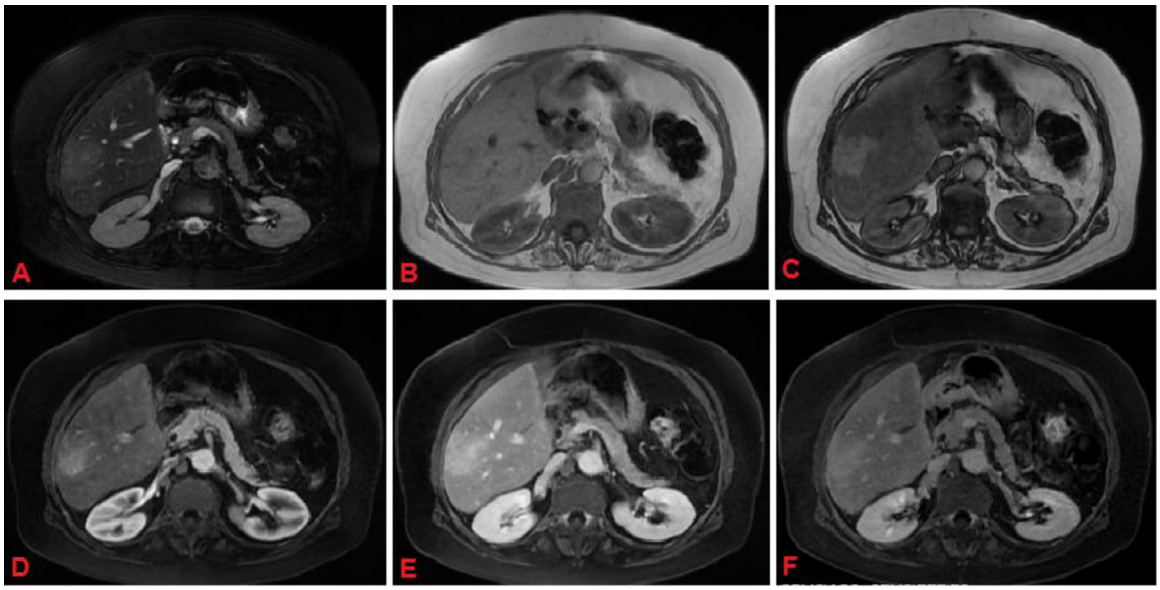

Fig. 2 – Hepatic MRI shows the lesion in right lobe. Note minimal high signal on T2-weighted images (A) and iso intensity on in phase T1 (B). On opposed phase T1 images (C), the lesion out stands with diffuse signal drop surrounding liver parenchyma due to steatosis. Also note enhancement pattern in early arterial phase (D), portal (E), and delayed (F) phase.

dothelioma 2 years ago. She had no other known comorbidities. She presented with elevated liver enzymes in her follow-up laboratory studies. Her laboratory data were aspartate transaminase (AST): 113 IU/mL (reference interval: up to 31), alanine transaminase (ALT): 292 IU/mL (reference interval: up to 32), alkaline phosphatase (ALP): 285 IU/mL (reference interval: 70-306), total bilirubin: 11.5 mg/dL (reference interval: 0.1-1.2), and direct bilirubin: 6.5 mg/dL(reference interval: up to 0.3).

On the liver ultrasonography, the liver showed increased echogenicity suggestive of fatty liver grade 2. There was also a hypoechoic mass in segment 6 of the liver measuring, 4.8 × 3.1 cm. Focal fat sparing and liver mass were the 2 most probable differential diagnoses suggested for the mentioned lesion. In order to evaluate the lesion more precisely, an abdominopelvic CT scan and dynamic liver MRI were performed, in which a 51 × 34 mm lesion with ill-defined borders and enhancement in early arterial phase was detected in segment 6 of her liver beside fatty liver infiltration (Fig. 1). On MRI, the lesion shows minimal high signal on T2-weighted images and iso intensity on in phase T1 and precontrast fat sat T1-weighted images. On opposed phase T1 images, the lesion out stands with diffuse signal drop surrounding liver parenchyma due to steatosis. On postcontrast images, the lesion depicts arterial hyper enhancement, which persists to delayed phase (Fig. 2). Focal fat infiltration was in the differential diagnosis but enhancement pattern and mass like morphology of the lesion necessitate biopsy.

The lesion was biopsied and sent to the pathology department. Microscopic examination of the specimen revealed liver

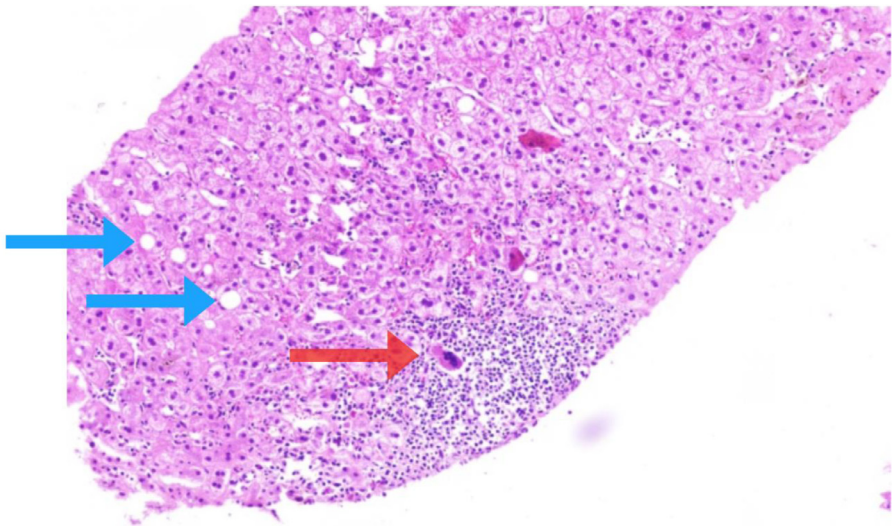

Fig. 3 – Liver tissue with focal extramedullary hematopoiesis. The red arrow shows a megakaryocyte surrounded by mixed myeloid and lymphoid cells. Scattered hepatocytes with fatty change (macrovesicular type) are also noted (blue arrows). H&E. x100.

tissue with preserved architecture and unremarkable portal tracts. Minimal lymphocytic infiltration was noted in some portal tracts along with equivocal endotheliitis and bile duct damage. Mild macrovesicular steatosis involving about 10% of lobular area was detected in the liver parenchyma. Sinusoidal congestion and intrasinusoidal infiltration of mixed inflammatory cells including some multinucleated giant cells (most probably megakaryocytes), T and B lymphocytes (CD3 and CD20 positive lymphocytes) were identified (Fig. 3) Multiple bile plugs were seen. Trichrome and orcein stains demonstrated no significant fibrosis. Rhodanine, Perl's iron, and PAS-D stains were all negative. C4d stain revealed minimal nonspecific staining in few portal tracts. The pathologic examination was suggestive of focal EMH.

It was concluded that the rise detected in our patient's liver enzymes was due to mild graft rejection. The mass detected in her liver was finally diagnosed as focal EMH.

Discussion

EMH presenting as a focal liver mass is a relatively rare clinical scenario [2]. It has been described to be hypoechoic or of mixed echogenicity on ultrasonography and usually hypodense or heterogeneously hypodense on unenhanced CT scan [2]. Our patient's liver lesion was also hypoechoic on ultrasonography and it enhanced on contrast-enhanced CT scan.

Our patient is a very rare case of post liver transplantation EMH presenting as focal liver mass. She did not have any underlying disease associated with ineffective hematopoiesis or hemolytic anemia to justify the presence of a focal EMH mass in her liver. Transient intrahepatic hematopoiesis has been reported in liver transplant patients [3,4]. However, focal EMH mass lesion in the transplanted liver is a very rare phenomenon. In a retrospective study of 312 liver transplant patients, liver aspirates of 39 patients (12.5%) showed signs of intrahepatic erythropoiesis. Nineteen patients had early postoperative intrahepatic erythropoiesis occurring in the first 3 postoperative weeks. The remaining had late postoperative intrahepatic hematopoiesis with a mean presentation time of 30-70 days postoperation. They hypothesized that this hematopoietic phenomenon can be a physiologic response in the transplanted liver [4]. Different theories including regenerative liver parenchymal activity and complex immune reactions in the allograft liver [4] as well as low hematocrit levels [3] have been proposed to play a role in post-transplantation intrahepatic hematopoiesis.

In a large case series study, it has been proposed that patients with idiopathic EMH most probably would not show any occult malignancy or disease in their primary and follow-up studies [5], so simple follow-up of our patient seemed a reasonable decision.

Authors' contribution

Study concept and design: **Faeze Salahshour, Niloofar Ayoobi yazdi**

Acquisition of data: **Ghazale Tefagh**

Analysis and interpretation of data: **Ghazale Tefagh, Faeze Salahshour**

Drafting of the manuscript: **Ghazale Tefagh, Faeze Salahshour**

Critical revision of the manuscript for important intellectual content: **Faeze Salahshour, Niloofar Ayoobi yazdi, Nahid Sedighi**

Administrative, technical, and material support: **Nahid Sedighi, Farid Azmoudeh ardalan, Masoume Safaei**

Study supervision: **Faeze Salahshour, Niloofar Ayoobi yazdi**

Patient consent

Written informed consent for publication has been obtained from the patient.

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