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A case of liver rupture in a patient with focal nodular hyperplasia at 33 weeks of gestation: A multidisciplinary management

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Dear editor,

Focal nodular hyperplasia (FNH) is the second most common benign hepatic lesion in adults. Etiology is unknown and it is often diagnosed incidentally in women of childbearing age. The disease has minimal risk of malignant degeneration or rupture.[1] It is often asymptomatic although it may manifest as abdominal pain or a palpable mass when the diameter exceeds 10 cm.[1–3] Despite the disease's predilection for females, there's no evidence that female hormones alter the course of FNH; several studies conducted on pregnant patients with FNH showed no modification in tumor size.[4–6]

In this article, we present the rare case of a hepatic rupture at 33 weeks of gestation in a patient with FNH.

A 29-year-old nulligravid female patient presented to the emergency department for a face edema and a diffuse rash consistent with urticaria. Her medical history consisted of urticaria and eczema, with a body mass index (BMI) of 33. Lab work showed elevated transaminases 2 times the normal level, gamma-glutamyl transferase (GGT) 67U/L and alkaline phosphatase (ALP) 129IU/L. C-reactive protein (CRP) was negative. She was treated with corticosteroids and antihistamines and discharged home with a prescription for hepatic ultrasound for further evaluation of the abnormal liver studies.

Hepatic ultrasound revealed diffuse steatosis and a 5 cm hepatic mass. The gastroenterology department was consulted, and they advised a hepatic MRI. MRI with hepatobiliary contrast agent was done and showed diffuse hepatic steatosis with multiple intrahepatic lesions, the largest in the left lobe measuring 57 mm, consistent with a diagnosis of focal nodular hyperplasia. A liver biopsy was recommended.

2 weeks after diagnosis of the lesion, the patient got pregnant. She presented to the gynecology ward for pregnancy follow up. Gastroenterology team was consulted, and they advised monthly check ups and close monitoring, with a follow up MRI in 3 months. Monthly lab results were in the range of normal, and hepatic MRI findings were unchanged. Obstetrical follow up was in the range of normal, with the exception of gestational diabetes, managed with diet.

At 33 weeks of gestation, the patient was brought to the gynecology ward for severe diffuse abdominal and pelvic pain of acute onset, 2 hours prior to hospitalization. Upon arrival, the patient was conscious and

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reactive. Blood pressure was 120/70 mmHg, pulse 120 bpm, temperature 36.1 °C. Physical exam showed soft abdomen with diffuse tenderness. Non stress test (NST) revealed fetal bradycardia of 50, not responsive to external stimuli. Decision was taken to proceed with urgent C-section for non-reassuring fetal heart. After a Pfannenstiel incision under general anesthesia, the gynecologist noticed 500 ml of blood clots with intact uterus. Fetal extraction was done with Apgar of 2 at 1 min and 0 at 5 min. Pelvic examination concluded non pelvic bleeding. The general surgeon on call was notified. He proceeded with midline incision. Exploration of the abdomen revealed an abundant hemoperitoneum with fresh blood and clots mainly in the left upper quadrant. After aspiration of the hemoperitoneum, a tumoral process was found on the left liver lobe and an ulceration of 15 cm near it with active bleeding. After manual compression for 30 min, packing of the liver with 12 packs was done and the decision was taken to monitor the patient with hepatic CT angiography and unpack if no active bleeding was observed. 2 packed red blood cells were transfused, hemoglobin was maintained at 11.4 g/L. CT scan repeated after several hours showed a 74 mm left lobe mass with no major active bleeding. Close monitoring was maintained for 48 hours. Unpacking was done, there was no active bleeding. Patient was extubated and monitored for 10 days in ICU. MRI was repeated after 5 days, it showed regression of the mass size to 50 mm (Fig. 1). She was discharged home with weekly follow up and an MRI after 3 months. The pathology result confirmed the diagnosis after 3 weeks, stating that the morphology and immunoprofile were suggestive of focal nodular hyperplasia.

Focal nodular hyperplasia (FNH) is the second most common liver disease after hemangioma. It affects female patients in the 4th and 5th decades, in 80%–90% of the cases.[1] The disease is mainly asymptomatic, and diagnosis is often incidental upon investigations for other pathologies. It may manifest as a palpable mass or a dull abdominal pain, especially in the presence of large nodules.[1–3] FNH has little or no potential for malignant degeneration, and intralesional hemorrhage is found in only 3% of cases with FNH.[7] Macroscopically, lesions appear as solitary nodules, well limited, nonencapsulated, ranging between a few mm and more than 10 cm, with a 'central scar' consisting of mature collagen, fibrous septae, veins and arteries.[1,3]

The diagnosis of FNH relies on imaging and histopathology; lab work is often normal.[3] In this case, the rise in the liver enzymes can be explained by liver steatosis, possibly due to the high BMI. Sonographic features on abdominal ultrasound consist of a hypoechoic or isoechoic nodule in 80% of cases with a feeding arteriole developing centripetally.[7] Due to the resemblance of FNH to hepatic adenoma on ultrasound, many scholars consider the MRI as the modality of choice for diagnosing FNH.[8] MRI features include isointense or hypointense nodule on T1-weighted MRI, and isointense or mildly hyperintense nodule on T2-weighted images.[7] Imaging may diagnose up to 90% of FNH, while 10% of the cases may still need histology for confirmation and to rule out hepatic adenoma or hepatocellular carcinoma.[1]

The differential diagnosis of FNH is mainly hepatocellular adenoma. Hepatocellular adenoma is usually symptomatic, with lesions ranging from 1 cm to 30 cm. It is associated with oral contraceptive pills (OCPs), and discontinuation of hormonal treatment has been shown to effectively reduce adenoma size. Diagnosis is based on MRI enhanced with gadobenate dimeglumine or gadoxetate disodium which can be very effective in differentiating hepatocellular adenomas from FNH and other lesions.[9,10]

The female to male ratio of FNH is 8:1, with increased incidence in the childbearing age.[11] This has inspired a lot of scholars to search for a causative factor related to female hormones. Although it has been known that oral contraceptive pills (OCPs) might affect the occurrence and progression of the disease, recent studies found no evidence to support this statement and discontinuation of OCPs after diagnosis of FNH is not indicated.[3–5,12] As for the course of FNH during pregnancy, few articles have discussed this subject. In a study by Mathieu et al. (2000), 12 patients were monitored during pregnancy and MRI showed no modification in the size of nodules (nodules range between 20 mm and 105 mm).[4]

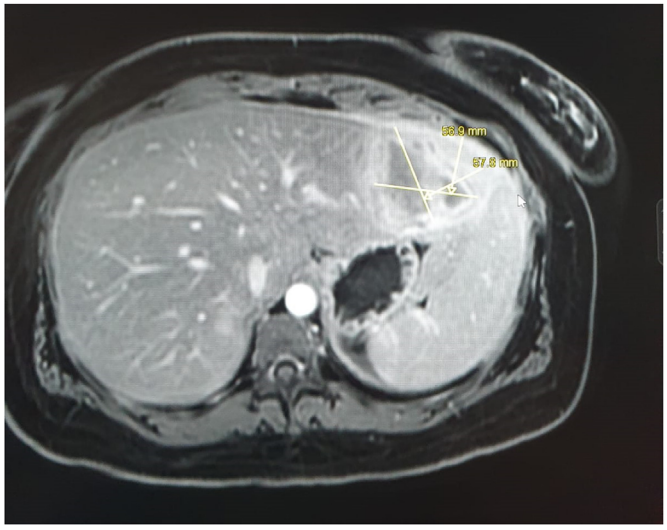

Fig. 1. Liver MRI showing regression of the mass size.

10 patients were also monitored during pregnancy in a study by Weimann et al. (1998) and showed no modification in the size of nodules.[6] In a study conducted by D'halluin (2001) on 44 patients, six patients became pregnant and 6 patients went into menopause during follow up, the lesion remained stable in 3 and 4 patients respectively.[5] The literature notes two cases of liver rupture associated with FNH during pregnancy: a 29-year-old patient with a rupture at 37 weeks of gestation, and a 23-year-old patient with a rupture at 39 weeks of gestation.[13,14] Both patients were not known to have FNH before pregnancy, and diagnosis was made after rupture.

As for the management of focal nodular hyperplasia, the European Association for the Study of the Liver (EASL) recommends a conservative approach due to the rarity of complications, and no follow up imaging is required in asymptomatic patients. Surgical management is reserved for patients with pedunculated, expanding and exophytic lesions. Pregnant patients with FNH are also exempt from follow up imaging.[15] On the other hand, hepatocellular adenomas tend to increase in size during pregnancy and conception is highly discouraged when tumors are larger than 5 cm.[10]

Studies investigating the course of pregnancy in patients with focal nodular hyperplasia are minimal, and several cases of liver rupture have been described. Further studies are needed to assess the influence of female hormones and pregnancy on the disease, and to set proper management and adequate follow up.

Ethics approval and consent

Approval was obtained from the local ethics committee.
Patient or study participant consent for publication.

Conflict of interest

The authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest, or non-financial interest in the subject matter or materials discussed in this manuscript.

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