

“Double Trouble”: Preoperative Embolization in a Case of Double Rupture of Hepatic Hemangioma during Pregnancy

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J Gastrointestinal Abdominal Radiol ISGAR 2019;2:64–68

Abstract

Keywords

- ▶ pregnancy
- ▶ cesarean section
- ▶ embolization
- ▶ hepatic hemangioma

Hepatic hemangiomas are the most common benign space-occupying lesion of the liver. These lesions are associated with the gestational age of the patient. They can easily be missed and can rarely present with complications. Spontaneous rupture of hemangioma is a rare occurrence. Preoperative arterial embolization of hemangiomas plays a vital role to reduce intraoperative bleed during surgical resection. Here is a case of a young pregnancy presenting with a complicated outcome.

Introduction

Hepatic hemangiomas are the most common benign space-occupying lesion of the liver.¹ They are usually small and asymptomatic; however, they can develop a complication such as spontaneous rupture that can go completely unnoticed, especially in the background of a gravid uterus. Preoperative arterial embolization of hemangiomas plays a vital role to reduce intraoperative bleed during surgical resection. Here is a case with a similar clinical history and its multidisciplinary approach in the management of the complication.

Case Presentation

A 28-year-old patient in term gestation presented with an acute onset of abdominal pain following which she was immediately shifted to the closest health care facility. On clinical examination, the attending physician felt that pain was not because of ongoing labor following which ultrasound and blood workup were performed. Blood evaluation was suggestive of a hemoglobin drop (10–6 g%), and portable ultrasound showed viable fetus; however, there was peritoneal free fluid with fluid showing echogenic internal content. A large focal lesion was seen in the left lobe of liver, which had a heterogeneous echotexture. Doppler was

not performed because of the patient's clinical condition. The patient was prepared for an emergency cesarean section with exploratory laparotomy. A healthy live fetus was delivered. On further evaluation, the peritoneal cavity contained a hematoma, and the liver showed a large lesion with a focus of bleed. The bleeder was surgically managed by ligating the point of bleed. Three points were ligated. After 24 hours, the patient's condition deteriorated with a further drop in hemoglobin concentration and oxygen saturation. The patient was intubated and shifted to a tertiary health care setup for further management. Further imaging was performed at our center.

Emergency computed tomography (CT) of the abdomen was performed (16 slice, GE Healthcare, Milwaukee, Wisconsin, United States). The scan was performed in the supine position in suspended respiration. Omnipaque (Iohexol 350, General Electric Healthcare Little Chalfont, United Kingdom) was used as intravenous contrast media. The Contrast was injected using a pressure injector through an 18-gauge cannula sited in the left upper limb vein. Patients underwent a dual-phase enhanced CT scan with the arterial phase taken at 15 seconds and the venous phase taken at 60 seconds. Technical parameters were set as pitch (1.2), collimation (arterial: 16 × 0.6 mm and venous: 24 × 1.2 mm), rotational time (0.6 seconds), kV and mA (120), and bolus trigger at descending aorta.

received

November 13, 2018

accepted after revision

December 2, 2018

DOI <https://doi.org/>

10.1055/s-0039-1677787

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Noncontrast CT showed enlarged left lobe with a large round lesion with areas of mixed density within, which was well demarcated from the liver and hyperdense intraperitoneal content suggestive of hematoma. On contrast-enhanced CT left lobe of the liver was enlarged and replaced with multiple lesions showing early arterial enhancement with contrast enhancement following the aortic enhancement pattern on successive phases (►Fig. 1). The largest lesion was found to measure 13.5 × 8 × 6 cm (AP × TR × CC) and showed peripheral nodular enhancement with central nonenhancing area suggestive of hemangioma (►Fig. 2). In view of the patient's clinical condition (postpartum day 2 with a low hemoglobin concentration), immediate surgery was deferred, and a two-step approach was contemplated by the surgical team. The first step was to perform a preoperative embolization and the second step to

perform a left lobe resection after the patient's condition was stabilized. Preoperative embolization was performed under mild sedation. Through a right femoral arterial access, the celiac and hepatic arteries were cannulated, and the angiographic run was acquired. Superselectively catheterization of the left hepatic artery branch was performed with a 2.7F microcatheter (Progreat, Terumo, Tokyo, Japan) (►Fig. 3), and suspicious foci of contrast staining was embolized to stasis using 300 to 500 μm polyvinyl alcohol microspheres (Cook, Bloomington, IN, USA) (►Fig. 4). Further embolization was not contemplated as the patient was planned for surgery, and the embolization was performed to attain clinical stability. Postembolization angiography showed stasis of flow with no more staining. Post-embolization, there was no further drop in hemoglobin concentration and the value stabilized at 7.4 g%. Postprocedure a second-step left lobe

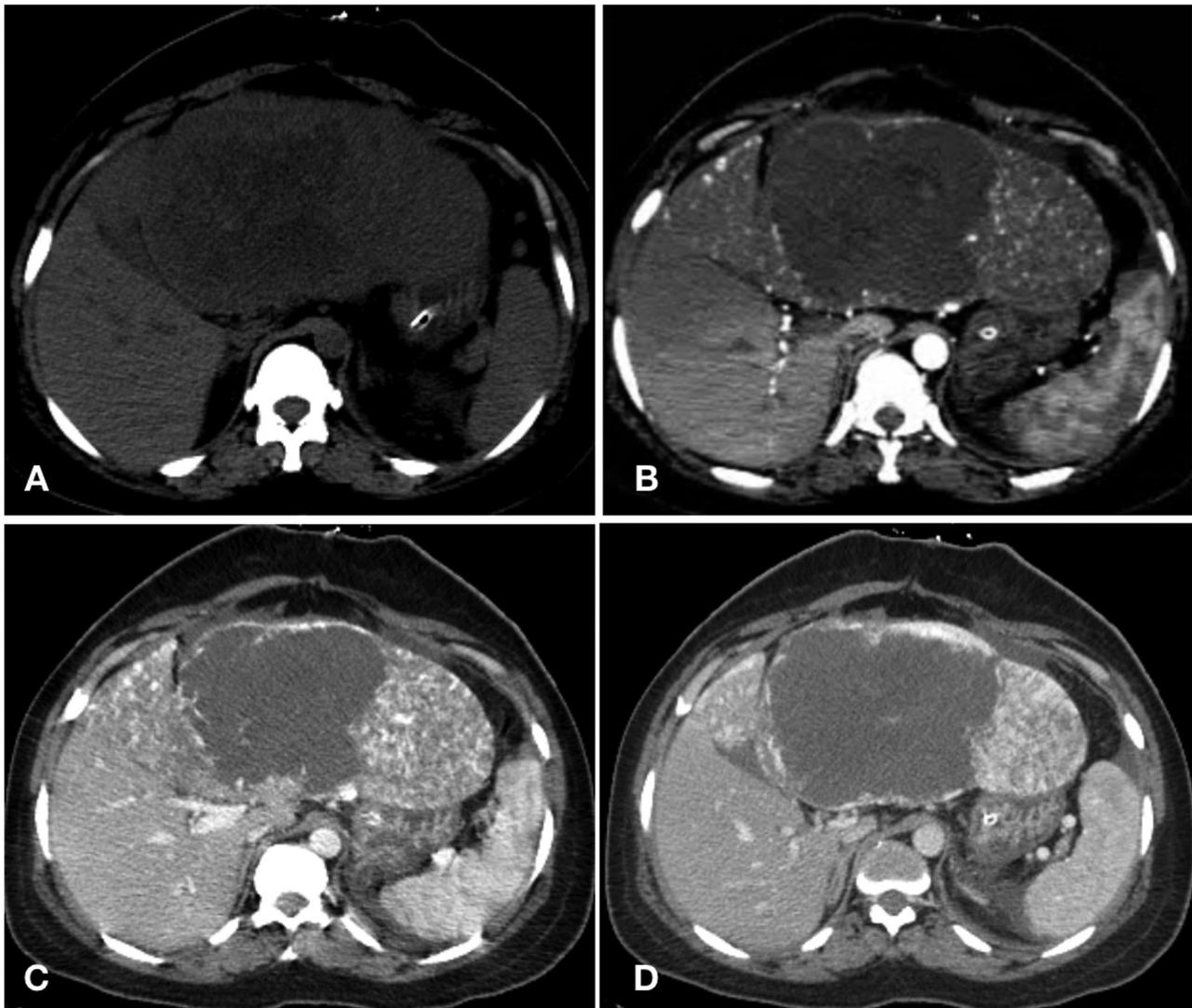


Fig. 1 (A–D) Axial images of contrast-enhanced CT of the abdomen. (A) Noncontrast scan shows bulky left lobe of liver with a large focal lesion showing some mixed density areas within. (B) Arterial phase shows multiple foci of intense arterial enhancement with a large lesion seen in segment 3. No active contrast extravasation was seen. (C, D) Portal and venous phase shows the lesion following the HU value corresponding to that of the aorta. The large segment 3 lesion shows peripheral puddling of contrast with the central nonenhanced area, which is suggestive of a hematoma.



Fig. 2 Contrast-enhanced CT of the abdomen. Coronal reformat image shows multiple liver lesions with a segment 3 lesion showing peripheral contrast puddling. No active contrast extravasation or capsular breach was seen.

resection was performed after 6 hours post-embolization. On the table, the surgeons found ease in resection due to the presurgical embolization as the blood loss was significantly low (30–40 mL). Post-surgery, the patient did well with no postoperative complication. Histopathologic evaluation of the specimen was suggestive of multiple capillary and cavernous hemangioma with many showing embolic particles within (► **Fig. 5**).

Discussion

Hepatic hemangiomas are the most common benign space-occupying lesion of the liver with an incidence rate of 0.4 to 7.3%.^{1,2} It is a vascular malformation of congenital origin with ectatic vessels that are either atypical or arranged irregularly. They can be either cavernous or capillary type. Lesions larger than 4 cm are known as a giant hemangioma. Hemangiomas that is, multiple hemangiomas are seen in up to 10% of cases.³ Hemangiomas are more common in females and have found to be more common during pregnancy and in patients taking exogenous estrogen or steroids. Hemangioma in pregnancy is different as its growth is triggered by various hormones.^{4,5} Lesion size is seen to be associated with the patient's gestational age.⁶ The progression and regression of the condition in the various group of patients have been poorly understood. In the vast majority of patients, the condition is asymptomatic and is incidentally detected on imaging. Giant hemangiomas can be symptomatic in 90% of cases.⁶ The patient presents with acute pain in the background of hemorrhage and thrombosis. Other complications include

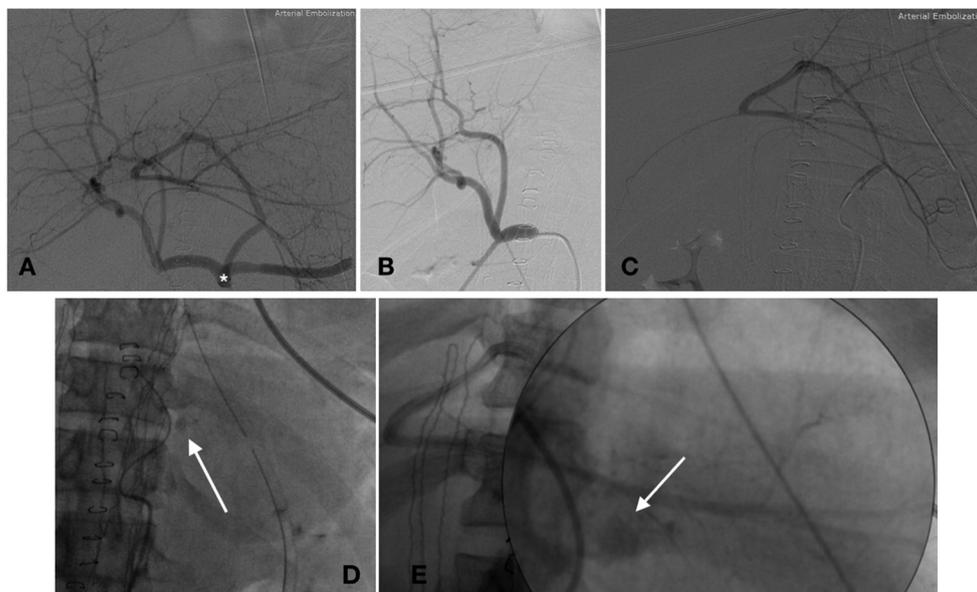


Fig. 3 (A–E) Conventional angiography. Celiac artery run showing replaced left hepatic artery, which is arising from the left gastric artery. **(B)** Common hepatic artery run. **(C)** Left hepatic run with the contrast blush. **(D)** Areas of contrast blush arising from the left hepatic artery vessels. **(E)** Magnified area of the contrast blush.

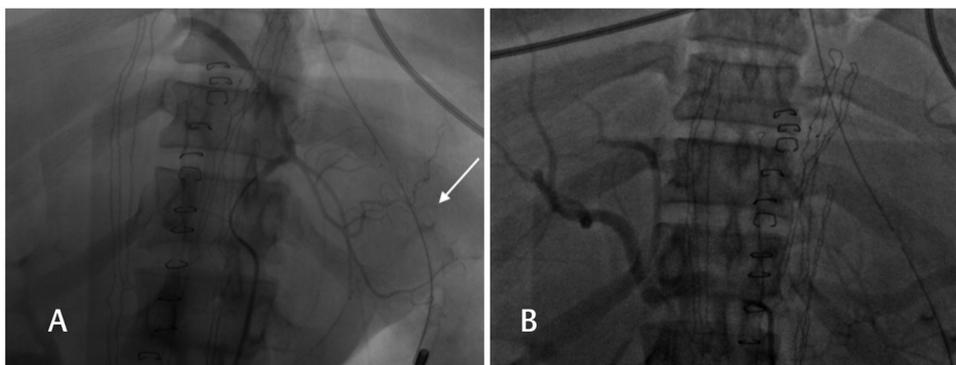


Fig. 4 (A, B) Postembolization images.

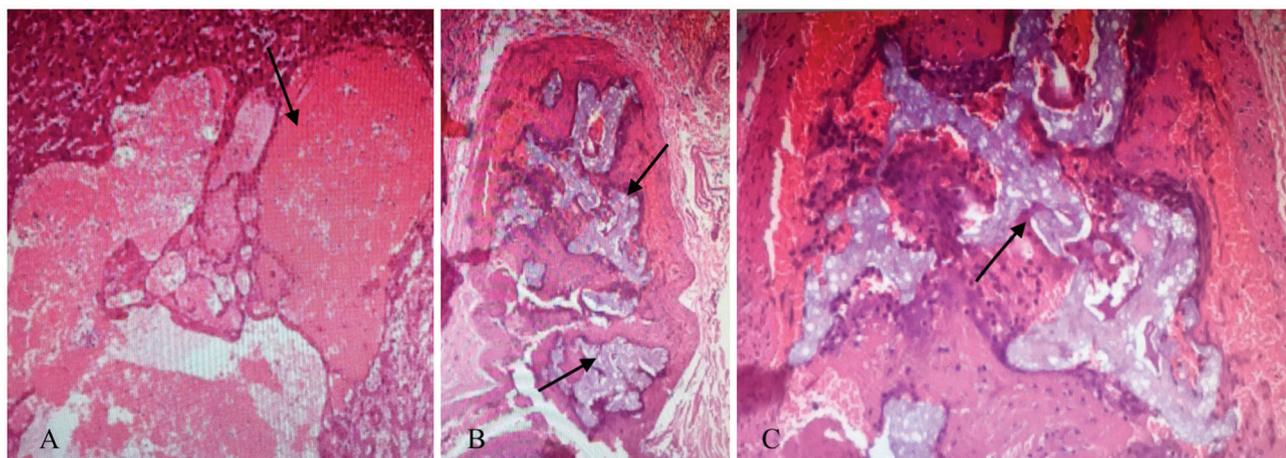


Fig. 5 (A–C) Histopathology of the resected liver specimen. (A) Large vascular spaces of varying sizes lined by flat endothelial cells, these spaces are separated by fibrous septa. (B, C) Few embolized spaces with the embolic material within the space.

anemia, thrombocytopenia, and hypofibrinogenemia.⁷ The lesion can easily be missed due to vague symptoms including epigastric pain, nausea, and vomiting. Spontaneous rupture is extremely rare and life threatening. Four types of rupture and bleeding have been described in the literature that includes (1) free rupture with hemoperitoneum, (2) intrahepatic hemorrhage, (3) double rupture, and (4) bleeding in the biliary tree.⁸ The hypothesis for spontaneous rupture is several and includes rupture secondary to the splitting of overlying liver parenchyma, necrosis secondary to rapid growth, and erosion of the hepatic vessels among a few causes.⁸

Various treatment option in treating asymptomatic hemangioma includes steroids, surgical resection, radiation therapy, and embolization.⁹ Worsening of symptoms, bleeding, or enlarging lesion size are indications for intervention. Various embolic agents can be used including PVA particles, gel foam slurry, iso-butyl cyanoacrylate, and coils mainly.¹⁰ Because of its permanent embolic nature, PVA particle was considered as an embolic of choice in our case.^{9–11} A review of the literature has revealed only a few cases of pregnancy associated with a complicated hemangioma.^{5,11–17} Transarterial embolization is a valid management tool due to evolving consumables. Presurgical embolization has shown very good outcome.^{18,19} Postembolic complication includes abdominal distention, fever, nausea,

vomiting, nontargeted embolization, hepatic dysfunction, and rarely bile duct injury. However, transarterial embolization is a very safe and effective option in patients with high risk for surgery.

Conclusion

This case offers several learning points: (1) Hemangiomas are common during pregnancy and presents with a vague atypical presentation, (2) difficulty in diagnosis during pregnancy and the limitation of imaging, and (3) treatment management of the condition depends on the patient's clinical status.

Key Points

1. Spontaneous double rupture of giant hemangioma during pregnancy is a rare occurrence that requires a multidisciplinary approach in the management of the condition.
2. Transarterial embolization is a safe, effective, minimally invasive procedure with a good outcome, especially in complicated hemangiomas.
3. Presurgical transarterial embolization is a very effective tool in the armamentarium of the interventional radiologist.

Conflict of Interest

None declared.

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