

# Gestational progression and blood supply assessment of a six-week primary hepatic pregnancy

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## Summary

**Background:** Primary hepatic pregnancy is a rare, but potentially fatal extra-uterine (ectopic) pregnancy. Most cases are missed in early pregnancy and frequently result in acute abdomen and hemorrhage during the first trimester. Only two cases reported were diagnosed prior to ten weeks of pregnancy. **Case Report:** The authors report one case of six-week primary hepatic pregnancy with timely diagnosis and subsequent evaluation by abdominal ultrasonography and describe the blood supply increasing of early hepatic pregnancy in 72 hours by color Doppler imaging. **Discussion:** Hepatic pregnancy progresses rapidly and the rupture risk increases day by day due to the rapid vessel-growth. When pelvic US fails to identify ectopic pregnancy, the present authors recommend a thorough abdominal US scan to expedite location of the gestational sac (GS) as early as possible. This is the mainstay of management to avoid serious potential complications. Lack of awareness about this condition may lead to missing or delaying diagnosis. **Conclusion:** It is very important that the clinician be aware of this condition with a high level of suspicion when pelvic US fails to locate the ectopic pregnancy.

**Key words:** Primary hepatic pregnancy; Ultrasonography; Ectopic pregnancy; Amenorrhea.

## Introduction

Primary hepatic pregnancy is a rare form of ectopic pregnancy that frequently results in acute abdomen and hemorrhage during the first trimester [1-5]. To the present authors knowledge, 23 cases have been reported in the English medical literature over the past 60 years [1-13]. As the primary screening method, US plays a central role in diagnosis of early ectopic pregnancies. Sonography can indicate the anatomic location of the gestational sac (GS), and color Doppler sonography is useful in determining whether the mass is vascular. However, only two cases presented as GS were discovered before eight weeks of gestation [2, 6], the others with live fetuses were diagnosed at 11-37 weeks of gestation. The present authors report the abdominal US diagnosis of a case of six-week primary hepatic ectopic pregnancy, and describe the gestation progression, as well as the blood supply increase within 72 hours by color Doppler imaging.

## Case Report

A 31-year-old woman, gravida 2, parity 1, presented with a three-week history of constant epigastric discomfort with nausea and was treated at the Department of Gastroenterology of the primary hospital. Gastroscopy showed superficial gastritis. Computed tomography diagnosed an abscess in the right liver. Symptoms worsened after a six-day symptomatic treatment, and hCG in serum was 49,195 mIU/mL (normally less than 3.0 mIU/mL) at 39 days of amenorrhea. She was referred to the pres-

ent hospital due to suspected ectopic pregnancy or choriocarcinoma and the presence of a liver mass.

The patient had had an appendectomy in 2004, gave birth via cesarean section in 2008, and had an intrauterine device placed following uterine curettage for a missed miscarriage in 2009. She reported a regular menstrual cycle of 32 days with normal menorrhea. On physical examination, there was tenderness in the right upper quadrant of the abdomen without rebound tenderness, no positive findings in the uterus or accessory reproductive organs, and no vaginal bleeding. Her serum hCG was 49,195 mIU/mL at 41 days of amenorrhea.

A transvaginal ultrasound (TVS) demonstrated a normal uterus with a T-type contraceptive ring. No gestational sac (GS), adnexal abnormality or pelvic fluid was observed. To identify signs of abdominal pregnancy or ectopic choriocarcinoma, an US examination of the abdomen was performed. The imaging displayed a 47×36-mm cystic mass located in the right posterior lobe of the liver near the branches of the vena portae. The wall presented as a hyperechoic ring with a honeycomb appearance and little non-echoic area with a 10-mm thickness. In the mass center, a 27×21-mm suspected GS was noted, including an amniotic sac, a yolk sac, and an embryo of 3 mm crown-rump length with no heartbeat. Color Doppler sonography detected diffuse blood flow signs around the mass (Figure 1) with parameters of the Doppler spectrum including a maximal velocity (Vmax) of 75 cm/s and resistance index (RI) of 0.36. The diagnosis was hepatic pregnancy. MRI was also performed, with findings in agreement with those obtained by US. She was treated with mifepristone orally (10 mg per day) with continuous monitoring of hCG, but her hCG levels continued to rise to 58,877 mIU/mL at 43 days of amenorrhea.

A subsequent US was performed to assess the progression of the mass. It showed the mass had migrated to Morison's pouch and increased to 54×48 mm. The honeycomb wall was thicker (14

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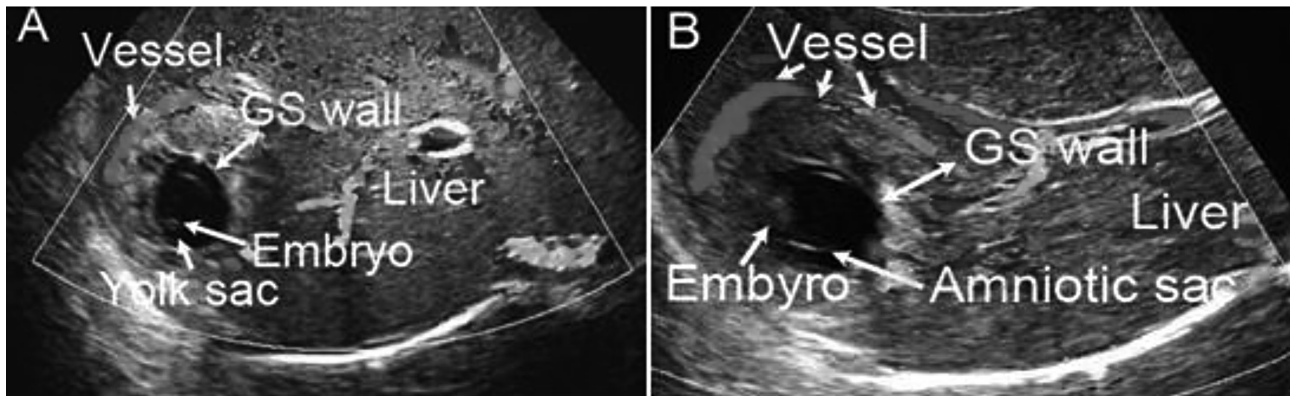


Figure 1. — Color Doppler sonography at 41 days of amenorrhea. (A) shows a round-shaped cystic mass located in the right posterior lobe of liver. A GS in the mass center includes an amniotic sac, a yolk sac, and an embryo, surrounding by a hyperechoic ring-shaped wall with a honeycomb appearance. Color Doppler sonography detects diffuse blood flow signs around the mass. (B) Sonography at 43 days of amenorrhea shows a thickened wall with a honeycomb appearance and indefinable embryo. Color Doppler sonography detects several new arteries and veins surrounding the mass. (GS wall: the thick wall of the gestational sac).

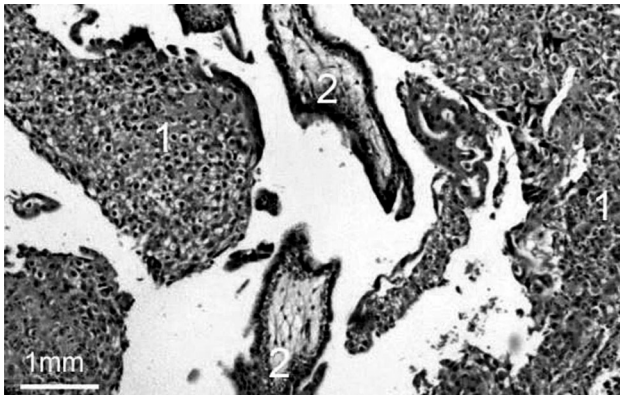


Figure 2 — Light microscopy (magnification  $\times 40$ ) shows an abundance of chorionic villi (marked as “2”) and trophoblast cells in the hepatic sinusoid (“1” represents a hepatic cell).

mm) with increased non-echoic area, and the diameter of the GS remained unchanged with a poorly defined embryo (Figure 1). There was a remarkable growth of new blood vessels. Color Doppler sonography detected several new arteries and veins with a diameter of approximately 4.0 mm surrounding the mass (Figure 1) and increased Doppler spectrum parameters: Vmax was 116 cm/s, and RI was 0.46. The US revealed high risk of hemorrhage due to the rapid growth and abundant vascularity of the mass. An emergency operation was required.

Hysteroscopy was performed under general anesthesia with the patient placed in dorsal lithotomy position. The uterine cavity was normal in appearance with a “T”-shape IUD malpositioned in its left branch. Oviduct apertures were identified bilaterally. No chorionic villi were found in decidual tissues. IUD removal and diagnostic uterine curettage were performed, with subsequent abdominal laparoscopic exploration. The uterus was tilted backward and affixed to the uterus-rectum fossa. The increased uterine size was equivalent to that at 40 gestational days. Bilateral adnexa were normal in appearance. Most of the omentum adhered to the parietal peritoneum. The liver and gallbladder also had normal appearance. A hepatobiliary surgeon was asked to attend the case immediately. A dominant mass with rich vascularity was observed

on the sixth segment of the right liver under abdominal laparoscopy. Uncontrollable bleeding occurred during the procedure, which was immediately converted to a laparotomy. A mass with an approximate 2-cm normal liver was resected with normothermic primary hepatic intermittent ischemia. Bleeding was stopped successfully, and the partial hepatectomy ended safely. Operative time was 27 minutes, the amount of bleeding during operation was 700 ml, and the volume of blood transfused was 3.5 units. The patient safely returned after surgery.

Subsequent growth of chorionic villi and trophoblast cells into the hepatic sinus was observed under a light microscope (Figure 2). The patient had a good postoperative recovery and discharged with no complications. The materials were obtained in the context of this report approved by Human Research Ethics Committee of The Second XiangYa Hospital.

## Discussion

The diagnosis of primary abdominal pregnancy is based on four criteria: (1) normal fallopian tubes, (2) no uteroperitoneal fistula should be demonstrable, (3) the pregnancy should be related exclusively to the peritoneum, and (4) the pregnancy should be recent enough to exclude the possibility of secondary implantation following primary nidation in the fallopian tube [12]. The present case fulfills these criteria.

The liver is an exceptionally rare implantation site for abdominal pregnancies. There are several factors that contribute to ectopic implantation. Some researchers believe that hepatic pregnancy may relate to IUD and oral contraceptive use, uterine cavity operation, and pelvic inflammatory disease [2, 4]. In the present case, the authors considered the following factors as potential causes of hepatic pregnancy. Firstly, an IUD created additional risk of ectopic pregnancy. Secondly, several previous surgeries resulted in substantial adhesion in the pelvic and abdominal cavity, which may prevent implantation of a GS. Additionally, the liver is the largest solid organ of the abdominal

cavity, contains capsula fibrosa, and is a favorable site with profuse blood supply and high suitability for the growth of an embryo. Fertilized eggs first attach to the capsula fibrosa. As the GS develops, chorion infiltrates the surface of the liver to meet the constantly increasing blood supply of the embryo [6]. In general, the GS ruptures due to the shortage of blood supply prior to 12 weeks gestation [6, 12]. It is reported that the most frequent implantation location is the right hepatic lobe, followed by the liver hilum [3, 5, 6], and then the caudate lobe [1] and the left lobe [4].

As occurred in this case study, it has been reported that common clinical manifestations of hepatic pregnancy include amenorrhea, vomiting, and abdominal pain. All cases presented in the literature had high hCG levels and no pregnancy identified in the uterus or pelvis. Several therapies can be used to safely treat hepatic pregnancy [7-11]. While hepatectomy has been the traditional treatment, the injection of methotrexate into the GS has proved to be a novel and useful method of treatment. In this case, oral mifepristone did not inhibit the progression of the mass, and hepatectomy was the only choice when hemorrhage occurred.

To the present authors' knowledge, no reports of color Doppler ultrasound results of vessel-growth during early hepatic pregnancy, which is not difficult to discover by ultrasound during the first trimester. With the early use of abdominal US, timely diagnosis can be made: an extrauterine GS can be detected in the liver as a round cystic mass with a hyperechoic ring-shaped thick wall and yolk sac and embryo inside [2, 7].

In the present case, the authors performed US serially and found that the mass increased rapidly with wall thickening and new vessel development. The thick wall presented as a hyperechoic ring with a honeycomb appearance, little non-echoic area, and blood vessels growing around the wall. The GS was located in the center of the mass. Color Doppler sonographic finding showed obvious revascularization. Both the diameter of the vessel and the maximal flow velocity substantially increased within 72 hours, although there was a blood supply shortage for the quickly growing GS and the embryo was damaged. Under light microscopy, the thick wall of the mass mainly consisted of trophoblastic cell and chorionic villi. Trophoblastic invasion was thought to be the usual cause of maternal bleeding and disruption of the implantation site. These changes, as demonstrated by serial US, indicated very fast progression of the mass with high risk of rupture and hemorrhage. Thus, early diagnosis and timely treatment were imperative.

However, hepatic pregnancies are often missed based on clinical findings during the first trimester [13]. Lack of

awareness about such a disease may be the main cause of missed diagnosis. Therefore, it is crucial that clinicians be aware of this condition and display a high level of suspicion when pelvic US fails to find the ectopic pregnancy. The present authors recommend a thorough abdominal US scan to locate the GS as early as possible. This is the mainstay of management to avoid serious potential complications such as severe intraperitoneal hemorrhage as a consequence of ectopic rupture.

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