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CASE REPORT

In Utero Sonographic Findings of Giant Hepatic Hemangioma and Associated Perinatal Complications: A Report of Two Cases

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It is uncommon to diagnose fetal hepatic hemangioma during the antenatal period. We describe herein two patients with a giant hepatic hemangioma detected antenatally, both with perinatal complications. In Case 1, a fetal intra-abdominal mass, measuring 63 mm × 50 mm × 74 mm, was observed below the right lobe of the liver, and the presumptive antenatal diagnosis of hepatic hemangioma was made at 37 weeks of gestation. Antenatal imaging suggested an intratumoral hemorrhage, but postnatal clinical findings refuted this diagnosis. However, progressive thrombocytopenia and coagulopathy were noted just after birth, resulting in the diagnosis of Kasabach—Merritt syndrome. In Case 2, our ultrasound examination performed at 40 weeks of gestation revealed a mixed solid and cystic hepatic tumor, measuring 99 mm × 54 mm. Further, antenatal sonography revealed cardiomegaly, increased descending-aorta velocity, atrioventricular valvular regurgitation, and a dilated inferior vena cava, suggesting high-output cardiac insufficiency. Giant hepatic hemangiomas can lead to severe complications such as cardiac insufficiency and Kasabach—Merritt syndrome, and these complications may occur during the fetal or early neonatal period. Detailed prenatal evaluation using fetal imaging and cord-blood sampling is important to determine proper antenatal management of patients with giant hepatic hemangiomas and to allow for prompt postnatal treatment.

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Introduction

Hepatic tumors account for approximately 5% of childhood neoplasms [1]. Infantile hemangioma is the most common primary hepatic neoplasm, followed, in order, by mesenchymal hamartoma and hepatoblastoma [2]. Small hepatic hemangiomas are usually asymptomatic and seldom require therapy. Giant hepatic hemangiomas, defined by a diameter of >4 cm, are rare but can lead to severe complications, including consumptive thrombocytopenic coagulopathy (Kasabach–Merritt syndrome) [3], intra-abdominal hemorrhage due to tumor rupture, and high-output congestive heart failure due to arteriovenous shunting [4]. It is uncommon to diagnose fetal hepatic hemangiomas during the antenatal period, but we encountered two such patients, with complications manifesting during the antenatal or early neonatal period.

Case Reports

Case 1

A 33-year-old primiparous woman was referred to us at 37 weeks and 6 days of gestation for evaluation of a fetal abdominal mass. Our ultrasound examination revealed a single fetus of a size consistent with her estimated date of confinement. A fetal intra-abdominal mass, measuring 63 mm × 50 mm × 74 mm, was observed below the right lobe of the liver. The mass contained cystic and solid areas, and a distinct border between the mass and the liver was not obvious. There was no evidence of hydrops. Fetal magnetic resonance imaging (MRI) demonstrated a mass, with low T1 and inhomogeneous high T2 signal intensity, originating from the right lobe of the liver (Fig. 1). The presumptive antenatal diagnosis was hepatic hemangioma. Six hours after the first ultrasound, the size of the fetal abdominal mass was re-evaluated and found to be 90 mm × 72 mm × 76 mm. Doppler imaging showed hypervascularity (Fig. 2). Increased systolic peak velocity in the middle cerebral artery (94 cm/s; 1.67 MoM) was also noted. The possibility of progressive fetal anemia due to intratumoral hemorrhage could not be excluded, and we decided to perform emergent cesarean delivery.

A female infant weighing 2536 g was delivered, with an Apgar score of 5 at 1 minute and an Apgar score of 7 at 5 minutes. Retractions and grunting respirations were noted in the newborn, but she did not need mechanical ventilatory support. After stabilization, the infant was transferred to the neonatal intensive care unit for further treatment. Her initial laboratory data showed thrombocytopenia (platelet count, $8.7 \times 10^4/\mu L$), and coagulopathy [prothrombin time percentage (PT%), 60%; international normalized ratio (INR), 1.34; activated partial thromboplastin time (aPTT), 58.1 s; fibrinogen, 143 mg/dL; fibrin degradation products, 76.2 μg/min; D-dimer, 36.5 μg/mL]. The patient was mildly anemic, with a hemoglobin level of 12.7 g/dL. As this value did not worsen, the prenatal suspicion of intratumoral hemorrhage was not borne out. Computed tomography examination on the 2nd day of life demonstrated an 8-cm mass emanating from the right lobe of the liver. After administration of intravenous contrast,

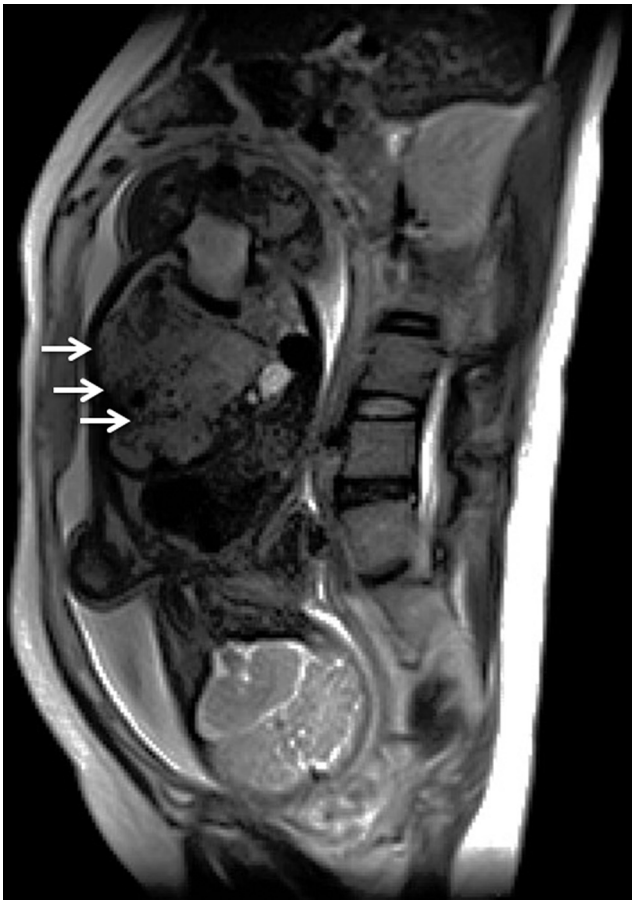

Fig. 1 T2-weighted coronal magnetic resonance imaging of a giant fetal hepatic hemangioma (arrows) at 37 weeks of gestation.

the lesion demonstrated peripheral enhancement with centripetal filling. The hepatic artery was markedly dilated (Fig. 3). Based on these findings, we confirmed the diagnosis of hepatic hemangioma. The infant gradually developed thrombocytopenia and coagulopathy. A blood sample taken on the 2nd day of life showed the following results: platelet count, $4.7 \times 10^4/\mu L$; PT%, 66%; INR, 1.32; aPTT, 90.4 seconds; fibrinogen, 92 mg/dL; fibrin degradation products, 92.1 μg/min; D-dimer, 46.2 μg/mL. These findings were compatible with disseminated intravascular coagulation, and we diagnosed Kasabach–Merritt syndrome. The infant's thrombocytopenia and coagulopathy were intensively treated with the administration of fresh frozen plasma, antithrombin, gabexate mesilate, and propranolol. The platelet count gradually stabilized and the coagulopathy resolved, and the baby was discharged home on Postpartum Day 67 with a platelet count of $22.1 \times 10^4/\mu L$. Over 5 months of follow-up and propranolol treatment, the neoplasm has grown smaller.

Case 2

A 44-year-old primiparous Japanese woman was referred to us at 40 weeks and 1 day of gestation because a fetal intra-abdominal mass had been identified on routine ultrasonographic examination. Our ultrasound examination revealed