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CASE REPORT

Spontaneous Regression of Infantile Hepatic Hemangioendothelioma

Wei-Ching Lin [1,2], Chien-Sheng Cheng [3], Chien-Heng Lin [4,5*]

[1] *Department of Radiology, China Medical University Hospital,* [2] *School of Medicine, China Medical University,* [3] *Department of Pediatrics, Jen-Ai Hospital,* [4] *Ho Meng Che Clinic, and* [5] *Department of Pediatrics, China Medical University Hospital, Taichung, Taiwan*

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Many primary hepatic tumors such as the rare pediatric neoplasms can be evaluated by sonography. Infantile hepatic hemangioendothelioma (IHH), the most common hepatic benign vascular tumor in infancy, has an excellent prognosis and spontaneous involution within 6—8 months in most cases. A 25-day-old baby girl was admitted to the hospital due to nonbilious vomiting for 2 days. Sonography showed multiple hypoechoic masses in the liver, and abdominal computed tomography demonstrated multifocal hypervascular lesions that were characteristic of IHH. The patient was then treated with supportive care and discharged in a stable condition without medication. Serial follow-up sonographic examinations showed that the size of the masses decreased gradually. At the age of 11 months, a follow-up sonogram confirmed complete resolution of IHH. Sonography is a useful and convenient method for diagnosis and follow-up of IHH. Long-term follow-up is necessary for IHH, even if the lesions are in regression, to watch for rare but possible malignant transformation.

Introduction

Primary hepatic tumors are rare in children and account for about 0.2—5% of pediatric neoplasms [1]. Sonography is a useful imaging modality to evaluate neonatal liver tumors. Infantile hepatic hemangioendothelioma (IHH), the most common hepatic benign vascular tumor in infancy, has an excellent prognosis and, in most cases, spontaneous involution within 6—8 months [2,3]. In the past, open liver biopsy and angiography were used most frequently to make the diagnosis. However, the sonographic feature of this condition has been described as multiple well-defined hypoechoic masses, and computed tomography (CT) demonstrated intense enhancement in these homogeneously hypodense masses after intravenous contrast agent administration. We present a case of IHH in which the

* Correspondence to: Dr Chien-Heng Lin, Ho Meng Che Clinic, Number 273, Shengli Road, Tanzi District, Taichung City 427, Taiwan.
E-mail address: lch227@ms39.hinet.net (C.-H. Lin).

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diagnosis was established with the use of noninvasive imaging techniques. Serial follow-up sonographic examinations were performed, and tumor regression without any medication was observed at the age of 11 months.

Case report

A 25-day-old baby girl presented with nonbilious vomiting for 2 days was transferred to our hospital. The infant was born at 37 weeks of gestation by normal vaginal delivery. Neither polyhydramnios nor abdominal mass was mentioned on prenatal sonographic examination, and there were no specific findings on the clinical examination at birth. On admission, her weight was 3.8 kg (25th and 50th percentiles) and length 57 cm (50th percentile). The heart rate was 130 beats per minute without murmur, and respiratory rate was 40 breaths per minute. On physical examination, abdominal distention and hepatomegaly were found 2 cm below the right costal margin, but there was no abdominal bruit on auscultation. No lymphadenopathy, splenomegaly, or cutaneous lesion was noted.

Initial laboratory evaluation showed that hemoglobin, hematocrit, platelet count, white blood cell count, and electrolytes were within normal limits. Plain films of the chest and abdomen revealed no specific findings. Abdominal sonography showed multiple hypoechoic tumors (the largest one was 3.8 cm in size) in the liver (Fig. 1A). On Doppler evaluation, blood flow without obvious shunting was noted within the tumors (Fig. 1B). Non-contrast-enhanced CT scan of the abdomen revealed diffuse, multifocal, homogeneous hypodense lesions of varying sizes in the liver (Fig. 2A); these lesions showed marked enhancement after the injection of contrast medium on arterial phase and remained mildly hyperdense in venous phase (Fig. 2B). Laboratory investigations of liver function later revealed that serum total bilirubin was 4.4 mg/dL (normal: <1.1 mg/dL), alanine aminotransferase 20 IU/L (normal: 5—40 IU/L), aspirate aminotransferase 33 IU/L (normal: 5—34 IU/L), and alpha-fetoprotein (AFP) 2946 ng/mL (normal: 30—5800 ng/mL). The multifocal tumors of vascular origin allowed the diagnosis of IHH to be made without the use of more invasive diagnostic procedures such as liver biopsy or angiography, which was considered unsafe due to a high risk of bleeding. Cardiac echogram showed no high-output congestive heart failure.

The patient was treated with supportive care and discharged in a stable condition without oral medication. The follow-up AFP level was 234 ng/mL (normal: 27—788 ng/mL) at the age of 3 months and 23 ng/mL (normal: 6—59 ng/mL) at the age of 11 months. A series of sonographic follow-up examinations (performed 1 month and 3 months, 6 months, and 9 months later) showed a gradual decrease in the size of the tumor and complete resolution at the age of 11 months (Fig. 3). The patient was completely clinically asymptomatic during follow-up.

Discussion

IHH, accounting for 2—3% of all pediatric neoplasms, is the most common vascular hepatic tumor in children [1]. Eighty-five percent of patients are diagnosed within the first 6 months of life, and female-to-male ratio varies from 1.3:1 to 2:1 [1—3]. Previous literature reported that the most common clinical presentations are asymptomatic hepatomegaly and abdominal mass (38—83%), followed by cutaneous hemangioma (11—66%) [1—3]. Congestive heart failure with a high cardiac output was observed in up to 50—60% of patients; however, this feature seems relatively rare (0—30%) in recent studies, especially in oriental children [3—6]. Our patient had a history of 2 days of vomiting prior to admission, which was considered due to overfeeding, excessive air swallowing, or viral acute gastritis and presumed not related to the hepatic tumor. The exact presentation of IHH in our patient was abdominal mass, which was found by a detailed physical examination after admission.

The initial investigation for diagnosis of IHH is sonography, which often shows a single or, more commonly, multiple iso- or hypoechoic masses in the liver. Although the lesion is often homogeneous, it may sometimes be

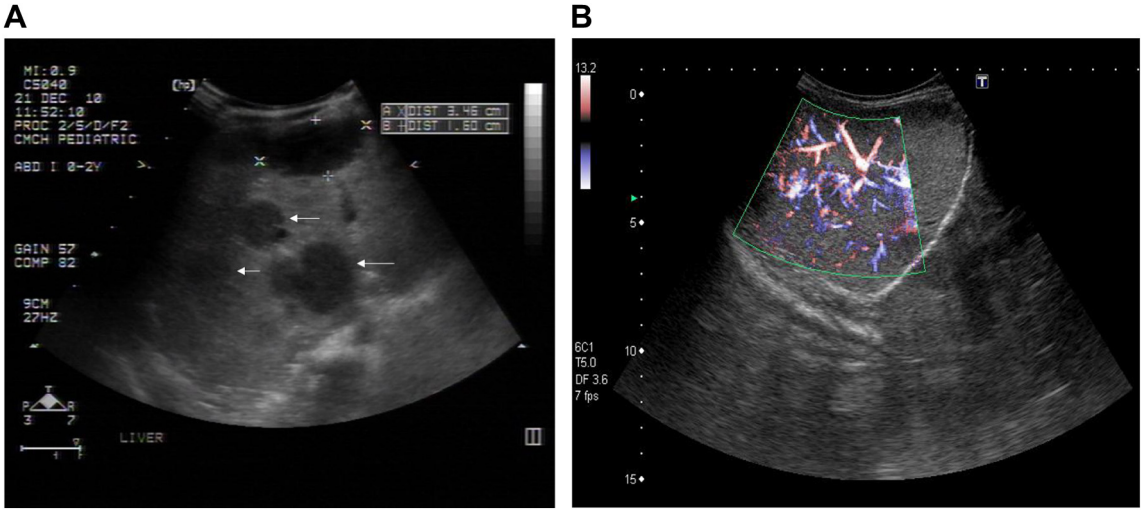

Fig. 1 (A) Abdominal sonography revealed multiple hypoechoic masses (white arrows) throughout the liver parenchyma. (B) Blood flow without shunting in the masses was noted on Doppler examination.