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Case Report

# Intraperitoneal hemorrhage in neonate with multifocal hepatic hemangiomas

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ABSTRACT

We describe a case of a 5-day-old male who presented with severe hemoperitoneum due to rupture of one of multiple hepatic hemangiomas, necessitating urgent embolization. Hepatic hemangiomas are common in the pediatric age group. The multifocal type typically presents shortly after birth, and have not been reported to bleed. The focal type is typically congenital with intratumoral bleeding described as a potential complication. We report a previously undescribed presentation of multifocal hepatic hemangiomas.



## Introduction

Hepatic hemangioma (HH) is the most common liver tumor in the first year of life, typically becoming apparent a few days to a few weeks after birth [1]. Three distinct subtypes of HH have been described: focal HH, multifocal HHs and diffuse HHs [2].

To the best of our knowledge, we present the first reported case of spontaneous rupture of multifocal HH resulting in severe hemoperitoneum necessitating embolization.

## Case Report

A 5-day-old male born at 39+4 weeks from an uncomplicated pregnancy was referred due to anemia of unknown cause, after child was noted to be jaundiced at a lactation clinic. His physical exam was unremarkable except for a markedly distended abdomen. No cutaneous vascular abnormality was present. The laboratory examination at presentation revealed severe anemia (hemoglobin: 51 mg/dL), and coagulopathy with thrombocytopenia (platelet count: 50 $\times$ $10^9$ platelets/L; prothrombin time [PT]: 20.7 seconds; international normalized ratio: 2.1; partial thromboplastin time: 36 seconds). The liver enzymes and bilirubin levels were within normal limits. Fibrinogen level was low (fibrinogen: <0.6 g/L [normal range 1.6-4.0g/L]).

At admission, abdominal ultrasound (US) showed multiple hepatic lesions of variable size, morphology and echogenicity, and marked abdominal ascites with low level echoes (Fig. 1). Echocardiogram was unremarkable. Due to concerns for active bleeding, an enhanced CT scan with arterial, venous and delayed phases of the abdomen was performed demonstrating multiple well-demarcated hepatic lesions showing peripheral enhancement with gradual filling-in, most in keeping with

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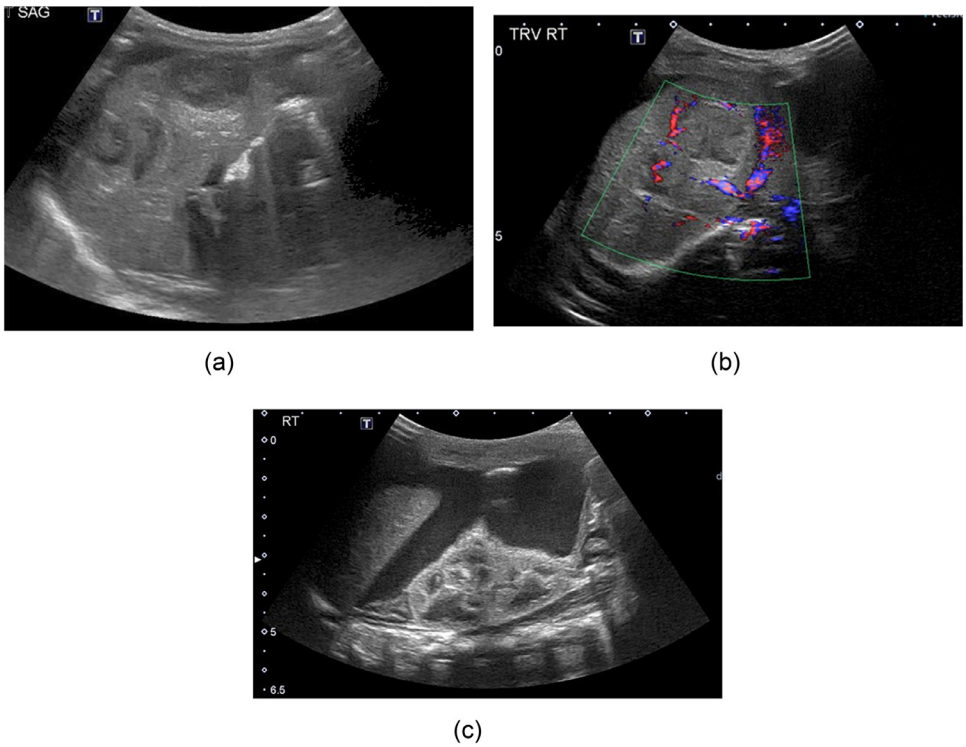


**Fig. 1 – US images show multiple liver lesions that vary in size and morphology in both hepatic lobes (a) demonstrating variable vascularity (b) and marked abdominal ascites with low level echoes (c).**

multifocal HHs. No arterial extravasation was identified. On the venous and delayed phases, extravasation was noted from a peripheral lesion in segment 2 compatible with active bleeding into the peritoneal cavity (Fig. 2). The patient was transferred to interventional radiology for urgent embolization. The common hepatic artery was accessed through a right brachial artery access. Digital subtraction angiography was performed showing diffuse abnormal vascularity consistent with the suspected multifocal HHs. The left hepatic artery (HA) was then selected with a microcatheter. No extravasation or shunting was noted on selective angiogram (as expected with venous phase hemorrhage). Polyvinyl alcohol particles of 150-250um were used to embolize the left HA. Repeat digital subtraction angiography demonstrated near complete occlusion of left HA (Fig. 3). Following the procedure, the child's hemoglobin stabilized with no need for additional transfusions.

Additional laboratory work was obtained to elucidate the nature of the liver lesions. Given the possible association of multifocal/diffuse HH with hypothyroidism, TSH was obtained at day 6 of life and was within normal limits (TSH: 2.12 mIU/L). However, a repeat study at day 14 of life showed elevated TSH levels (TSH: 11.29 mIU/L) with free T4 levels within normal limits (free T4: 26.7 pmol/L). Persistent elevated TSH with normal T4 levels was seen at the age of 8 weeks (TSH: 7.51 mIU/L, free T4: 19.2 pmol/L). and serial AFP measurements showed decrease in AFP level (10,400 ug/L and 9,945 ug/L at 2 and 4 weeks of age, respectively). Urine catecholamines were within normal limits.

A whole-body MRI was performed (coronal STIR images) showing the known multiple T2 hyperintense lesions within the liver, that could represent multifocal HHs, and a punctate, nonspecific lesion within the spleen that may represent a small hemangioma (Fig. 4).

Serial follow-up US studies up to 4 weeks of age showed no significant interval change in the multiple ill-defined liver lesions scattered throughout the liver. An US obtained at 8 weeks of life showed mild interval decrease of some of the hepatic nodules (eg, largest nodule largest diameter was 1.5 cm compared to 2.3 cm previously).

At approximately 3 weeks of age, the parents noted bright red soft papules on the right hip as well as on right scalp, that appeared clinically consistent with infantile hemangiomas. The skin lesions were then biopsied and were glucose transporter 1 (GLUT-1) positive capillary hemangiomas, consistent with infantile hemangiomas.

## Discussion

Focal HH, multifocal HHs and diffuse HHs vary in their clinical presentation, radiographic appearance, laboratory findings, natural history, and management approach.

Focal HHs in neonates are generally GLUT-1 negative and considered congenital HHs. They proliferate in utero and generally reach peak size prior to, or at birth [3]. The possible complications of a congenital HH include intratumoral bleeding, thrombocytopenia, hypofibrinogenemia, and high-output cardiac failure [4].

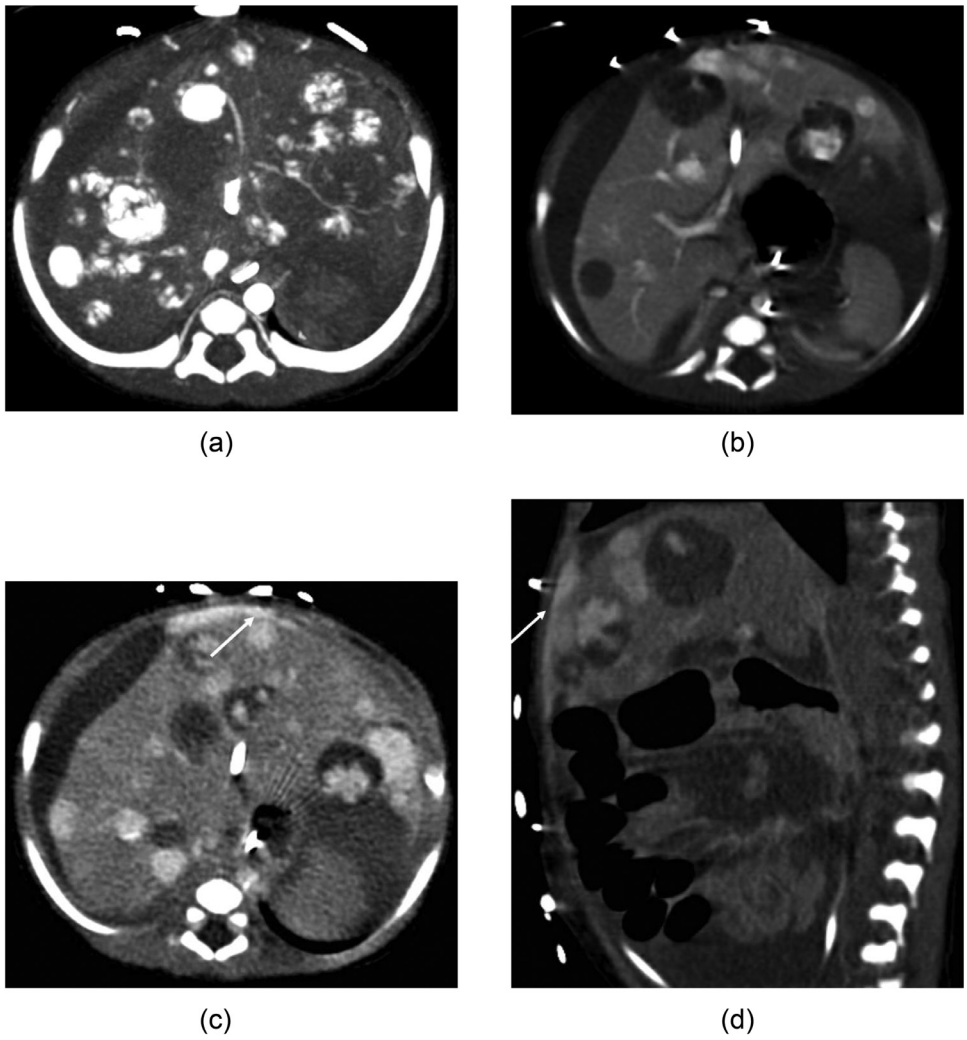


**Fig. 2 – CT at arterial (a), venous (b), and delayed phases (c-d) show multiple well-demarcated hepatic lesions of various size and enhancement times. Extravasation (arrows) of contrast material from an anterior lesion in the left lobe of the liver is evident (c-d) as well as the massive ascites (hemoperitoneum).**

Multifocal and diffuse HHs, like the more common cutaneous infantile hemangiomas, are GLUT-1 positive [2,4]. Most patients are asymptomatic, but some may present with significant arterio-venous shunting, respiratory distress, and high output cardiac failure. The diffuse form of HH may present with serious complications such massive hepatomegaly causing compression of the inferior vena cava and thoracic cavity, respiratory distress, abdominal compartment syndrome, and multiorgan failure or severe hypothyroidism due to the overproduction of type III iodothyronine deiodinase [1,2]. Symptomatic multifocal or diffuse HH are traditionally managed medically with steroids and propranolol. Patients with complicated clinical course are often referred for arterial embolization, surgical resection, or liver transplantation [1,5].

The differential diagnoses for neonatal HHs include metastatic neuroblastomas, hepatoblastomas, and mesenchymal hamartomas. Metastatic neuroblastomas, unlike HH, are associated with elevated levels of urinary catecholamines. Furthermore, imaging findings of additional sites of metastases or the primary tumor suggest the proper diagnosis. Hepatoblastomas rarely occur in the newborn and are distinguished by markedly elevated levels of alpha fetoprotein (AFP). Mesenchymal hamartomas of the liver, like HH, may also be found in the perinatal period, however, they usually appear as multicystic masses with enhancement of only the septa and solid portions [6].

We describe a case of multiple hemangiomas in the liver, presenting at 5-days of age with rupture and severe hemoperitoneum necessitating urgent embolization. Given the elevated TSH and thrombocytopenia, as well as the presence of cutaneous infantile hemangiomas, multifocal infantile liver hemangiomas is favored at this time. The only “gold standard" way to determine this would be to biopsy the lesions, however, such a procedure carries high risk of hemorrhage and is not felt to be clinically warranted.

The described case is unique for several reasons. Although bleeding of a HH (usually focal hemangioma) is relatively common, the hemorrhage is typically intratumoral, rather than

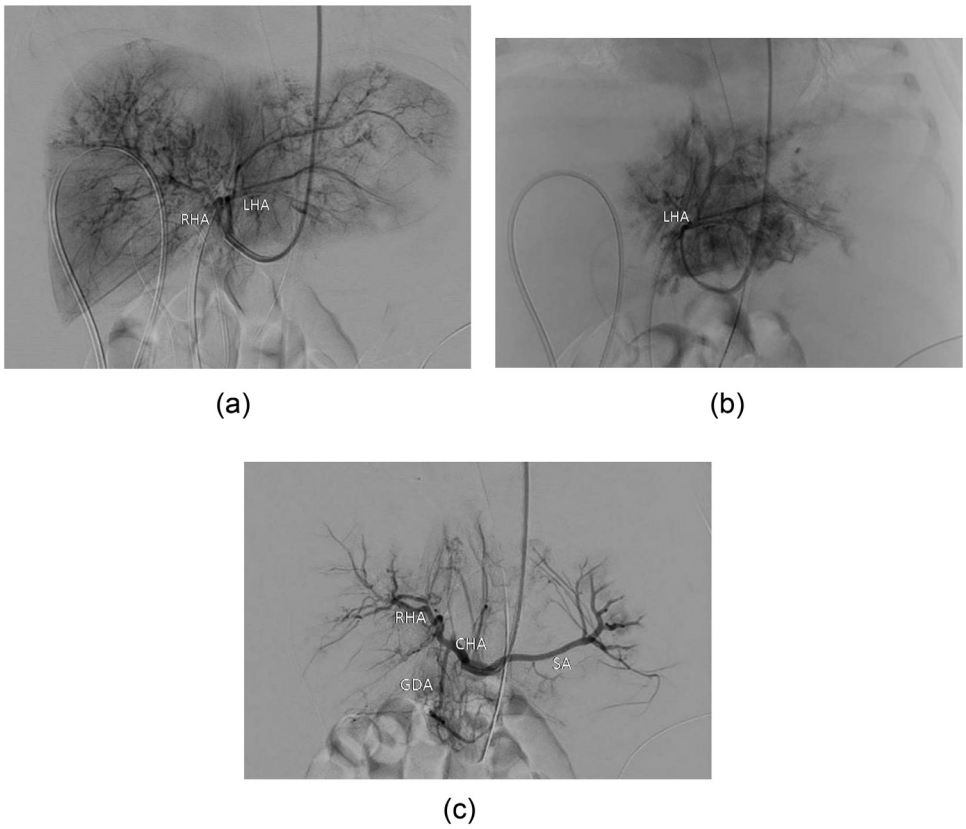


**Fig. 3 – Common hepatic digital subtraction angiography (DSA) performed demonstrating diffuse abnormal vascularity (a). A 2.4 French Progreat was used to access left hepatic artery (HA) and DSA repeated (b). No shunt or extravasation was noted. 150-250 um PVA particles used to embolize left HA. Repeat DSA demonstrated occlusion of the left HA (c). Splenic artery (SA), left hepatic artery (LHA), right hepatic artery (RHA), common hepatic artery (CHA), and gastroduodenal artery (GDA).**

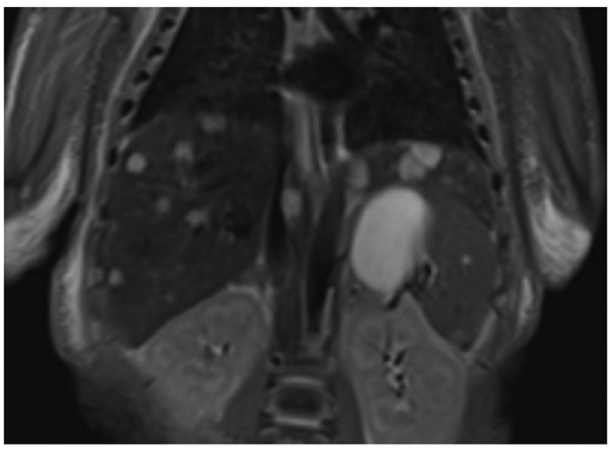

**Fig. 4 – Coronal STIR MRI image showing multiple T2 hyperintense lesions within the liver, that may represent hemangiomas, and a punctate T2 hyperintense signal within the spleen, is nonspecific but may represent a small hemangioma. Other organs were unremarkable.**

intra-peritoneal bleed. In addition, although embolization of HHs has been reported in the literature [2,7], it is mainly to manage shunts leading to congestive heart failure rather than acute bleeding. The very early presentation of multiple HHs has been described before, however, uncommon in the very early neonatal period [8].

This case describes a common diagnosis of multifocal HHs, with a rare acute and early presentation. Although a definite histopathological diagnosis was not obtained, the physical examination, laboratory work, and findings on imaging suggest the diagnosis. Embolization was clinically warranted regardless of the histopathological diagnosis, enabling stabilization and further management.

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