Idiopathic perinatal hepatic infarct as a cause of liver mass

Josep Marti[a], Anshu Trivedi[b], Ally Rosen[c], Ronen Arnon[a], Swan N. Thung[b] and Myron Schwartz[a]

We present the case of a 2-week-old male infant who presented with an asymptomatic liver mass and underwent surgical resection because of suspicion of malignancy after extensive radiological study with ultrasonography, computed tomography, and MRI. Pathological examination revealed a peripheral hepatic infarct with calcifications and a rim of peripheral organization suggestive of being at least 2 weeks old. This case reviews the scarce cases of hepatic infarct in newborns and highlights the fact that, although untreated perinatal hepatic infarction usually progresses to atrophy of the affected area with compensation by the unaffected liver, surgical resection remains an option in cases of complications or uncertain diagnosis. ***Ann Pediatr Surg*** **12:167–169 © 2016 Annals of Pediatric Surgery.**

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[a]Recanati-Miller Transplantation Institute, [b]Division of Liver Pathology, The Lillian and Henry Stratton - Hans Popper Department of Pathology and [c]Department of Radiology, Icahn School of Medicine at Mount Sinai, New York, New York, USA

Correspondence to Josep Marti, MD, PhD, Recanati-Miller Transplantation Institute, Icahn School of Medicine at Mount Sinai, 1 Gustave L. Levy Place, PO Box 1104, New York, NY 10029-6574, USA
Tel: +1 212 659 8522; fax: +1 212 348 2474; e-mail: josepmartis@yahoo.es

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Introduction

Hepatic masses are uncommon in the perinatal period but they are associated with significant morbidity and mortality in affected patients [1–4]. We report the case of an unusual benign entity to raise awareness about the importance of the newborn's clinical history details when performing differential diagnosis of neonatal liver masses.

Clinical history

The patient was a 2-week-old male who was transferred to our institution for evaluation of a liver mass diagnosed by ultrasound, which was performed because of findings of hypospadias and a right neck skin tag. The patient was one of the twins, born at 36.6 weeks of gestation via elective cesarean section with Apgar scores of 8 and 9. On physical examination, apart from the findings that led to the ultrasound study, jaundice was noted and the liver was palpable 4–5 cm below the right costal margin.

Repeat noncontrasted Doppler ultrasound performed at our institution demonstrated a mass in the posterior right lobe of the liver measuring $3.3 \times 1.9 \times 2.6$ cm with an ill-defined margin, heterogeneous echogenicity, and little vascular flow within the mass. Computed tomography scan showed a heterogeneous, predominantly hypodense mass, measuring approximately $3.0 \times 1.9 \times 2.4$ cm in the posterior right hepatic lobe (segments VI and VII) with peripheral and patchy internal enhancement on arterial phase imaging and with further filling on delayed phase images, suggesting the diagnosis of hemangioendothelioma with an area of necrosis. We subsequently performed MRI, which demonstrated a mass in the right posterior liver with hypointensity in T1 and hyperintensity in T2 (Fig. 1). Liver function tests showed normal aspartate aminotransferase, alanine aminotransferase, and alkaline phosphatase but elevated γ-glutamyl transpeptidase (412 IU/dl). Preoperative total bilirubin was 5.1 mg/dl, predominantly unconjugated; international normalized prothrombin ratio and serum albumin were normal, and α-fetoprotein (corrected for gestational age) was 290 000 ng/dl.

After discussion with the family, it was decided to proceed with surgical resection because of suspicion of malignancy, particularly in view of the elevated α-fetoprotein. Right hepatic lobectomy was uneventful; there was no evidence of extrahepatic disease. There were no postoperative complications and the patient was discharged on post-operative day 6. Pathological examination revealed a peripheral hepatic infarct with calcifications and a rim of peripheral organization with fibroblasts and granulation tissue (Fig. 2). Although the exact age of the infarct is difficult to predict, it appeared to be at least 2 weeks old because of the fact that it showed peripheral organization.

Discussion

Perinatal hepatic infarction is a rare occurrence that has been described in association with congenital infection, intrauterine asphyxia, placental thrombosis, umbilical venous catheterization [5], congenital heart disease [4,6], maternal oxytocin overdose [7], and twin-to-twin transfusions [8–11]. Hypotheses about the mechanism of infarction include embolization of a placental thrombus via the umbilical vein and liver ischemia due to vasoconstriction or hypotension; specifically in twins, a transfer of thromboplastin-rich material via the placenta/umbilical vein has been suggested [8]. Presentation of perinatal hepatic infarction ranges from an incidentally discovered asymptomatic finding to varying degrees of hepatic failure and portal hypertension, depending on the extent of infarction. In some cases, perinatal hepatic infarction has been associated with infarction of other organs including the brain [9].

The diagnosis of perinatal hepatic infarction is usually radiological, although some cases require confirmatory surgical exploration and pathological analysis. The most common initial study is ultrasound, which typically shows

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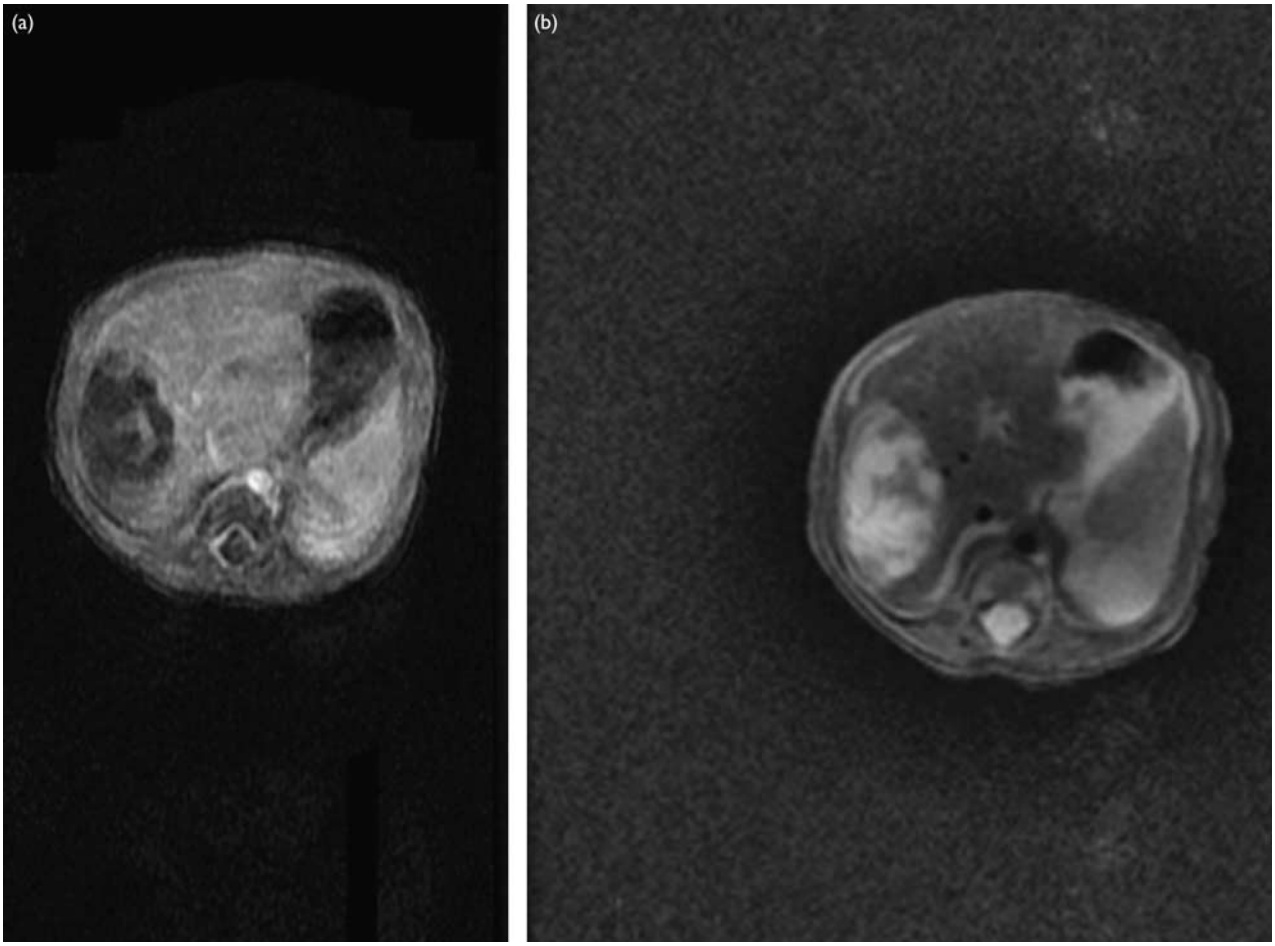
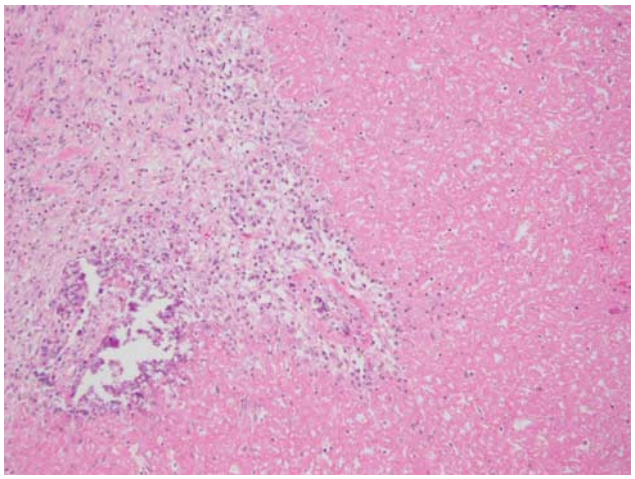
Fig. 1



MRI scans showing a mass in the right posterior liver with hypointensity in T1 (a) and hyperintensity in T2 (b).

Fig. 2



Edge of the liver infarct showing a rim of organization and calcification (hematoxylin and eosin, × 100).

a hypoechoic mass with or without a wedge-shaped distribution, depending on the affected vessel; flow within the mass is typically not observed on Doppler examination [9]. Computed tomography and MRI are the modalities of choice in the diagnosis of hepatic infarction, providing useful information regarding site, morphology, and extent of the lesion. Typically, a hypodense wedge-shaped lesion without contrast enhancement is seen, although rounded central or marginal lesions have also been observed [12–15]. Untreated, perinatal hepatic infarction progresses to atrophy of the affected area with compensation by the unaffected liver, provided that complications such as infection or hepatic failure do not alter its natural evolution; thus, when confidently diagnosed on imaging, close observation with supportive care is the treatment of choice [16]. As in our patient, interventional or surgical treatment is reserved to cases in which complications arise or the diagnosis is in doubt.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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