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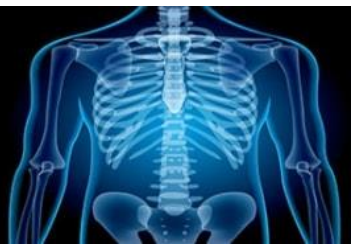

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Infantile hepatic hemangioma: A dual case study and review of literature

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Abstract
Infantile hepatic hemangiomas (IHHs), which are the most common infantile benign liver tumors shows a similar proliferative and involutional stages as infantile hemangiomas on the skin. IHHs are frequently asymptomatic, although some lesions can cause serious illness in the form of hypothyroidism, coagulopathy, and high-output congestive heart failure, which makes early detection as much as effective management crucial. The authors propose that as a first-line modality, ultrasound (US) is ideal because not only is it highly specific in its diagnostics, it is also highly effective in monitoring lesions involution, with computed tomography (CT) and magnetic resonance imaging (MRI) held in reserve in case ultrasound is not definitive. Through the analysis of the two cases below and a review of the literature, we aim to provide a detailed description of the multiple radiological aspects of this pathology, as well as an in-depth assessment of the available therapeutic approaches.

Keywords: Infantile hepatic hemangiomas, ultrasound imaging, pediatric vascular anomalies, therapeutic management

Introduction

Case reports

First Case

A 4-day-old female neonate was admitted for the management of an abdominal mass. She was born to non-consanguineous parents with no significant past medical history. The pregnancy was not followed up in a satisfactory manner although not complicated, with a resulting full-term birth with APGAR score 4/10/10; the neonate required mask ventilation at birth. There was no history of maternal infection.

Symptom development occurred on the date of admission with development of acute respiratory distress in terms of tachypnea with cyanosis that responded with oxygen. Clinical examination revealed a 5.5 cm mass in the right hypochondrium with extension into the right flank with collateral circulation. Initial labs reported a hemoglobin level of 7.2 g/dL and a platelet count of 30,000/mm³; these merited transfusion with packed red blood cells as well as with platelet concentrate. The level of serum alpha-fetoprotein (AFP), as well as beta-human chorionic gonadotropin (β-HCG), was reported as in normal ranges, and a low TSH level, indicating hypothyroidism. A big, lobulated, heterogenous abdominopelvic mass on US (Figure 1) with its vessels on Doppler revealed a mass that displaced both the above liver as well as kidneys laterally, with intimate contact with both aorta as well as with the inferior vena cava, with accompanying ascites. CT (Figure 2) reported a big mass in the liver that involved segments V, VI, VII, as well as VIII, which is seen with exophytic growth into the right iliac fossa. The mass has irregular contours and demonstrates heterogeneous, peripheral nodular enhancement following contrast administration, delineating areas of necrosis and containing peripherally arranged calcifications. The mass measures 71 x 86 x 100 mm and remains at a distance from the abdominal aorta and its visceral branches as well as from the iliac bifurcation. A small amount of intraperitoneal fluid is also noted.

Immunosuppressive therapy with Afinitor (mTOR inhibitor and antineoplastic agent) at 26 mg daily was initiated. Computed tomography scan as well as ultrasound (Figure 3) conducted a year following initiation of treatment revealed a significant reduction in the size of the necrotic hepatic mass estimated at 91%.

Later on, the patient underwent tumor resection with an uncomplicated postoperative course.

The histopathologic examination revealed a small-vessel vascular proliferation lined with non-atypical endothelium and filled with blood, in association with extensive areas of necrosis, calcifications, and hemosiderin deposits. These findings support the diagnosis of a remodeled hepatic capillary hemangioma.

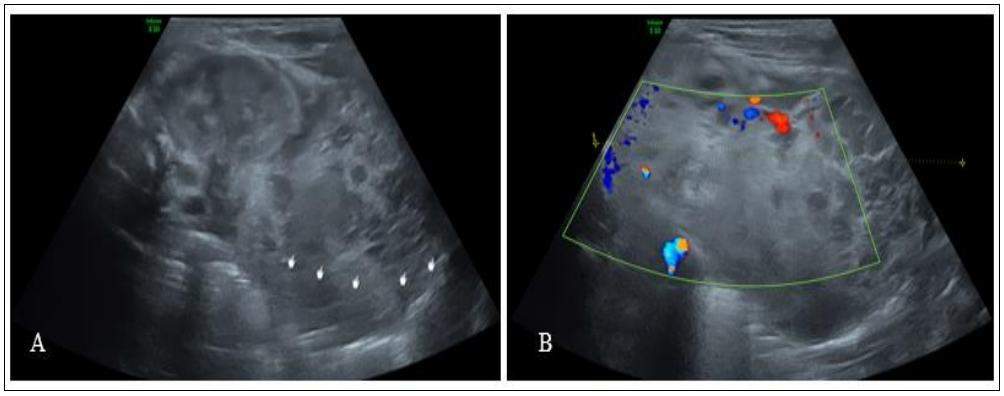

Fig 1: There is a large tissue mass, seemingly arising from the right liver, that is poorly circumscribed with irregular borders and a heterogeneous echotexture containing hypoechoic areas (Fingers) (A). The lesion is vascularized on color Doppler imaging (B) and measures approximately 11.5 cm at its greatest diameter.

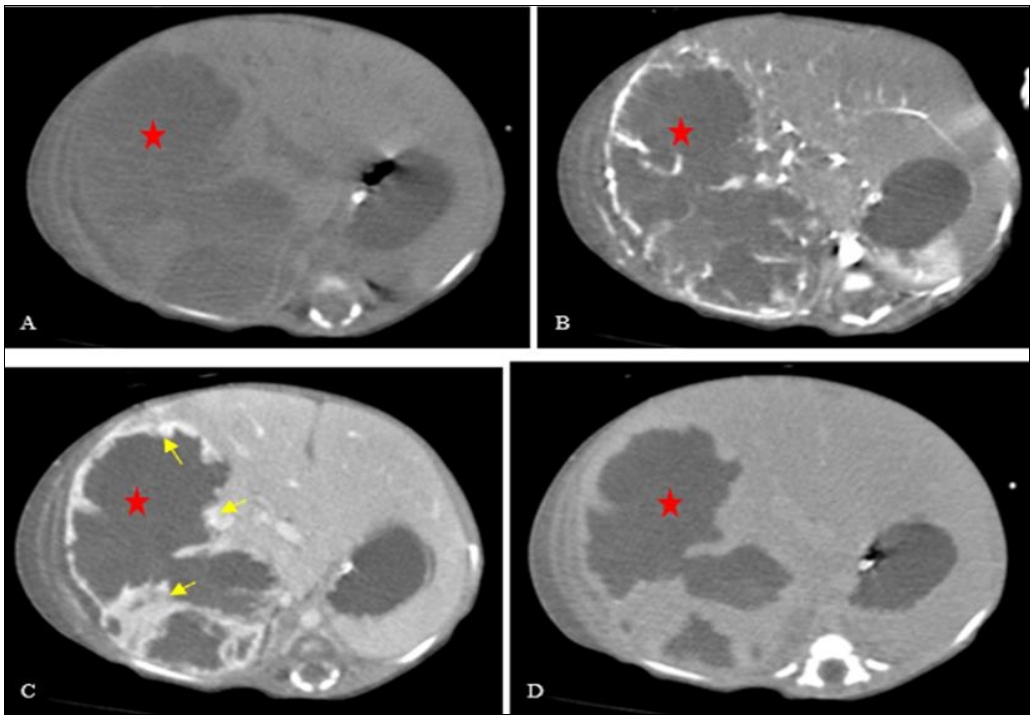

Fig 2: Axial sections through the abdominal region were obtained in the non-contrast phase (A) followed by arterial (B), portal (C), and delayed phases (D). These images demonstrated a large hepatic mass involving segments V, VI, VII, and VIII with an exophytic growth pattern extending to the right iliac fossa. The mass displayed irregular margins and heterogeneous, peripheral nodular enhancement after contrast injection (yellow arrow), delineating areas of necrosis (★). The lesion measured 71 x 86 x 100 mm, findings consistent with a large congenital hepatic hemangioma, likely with fissuring.

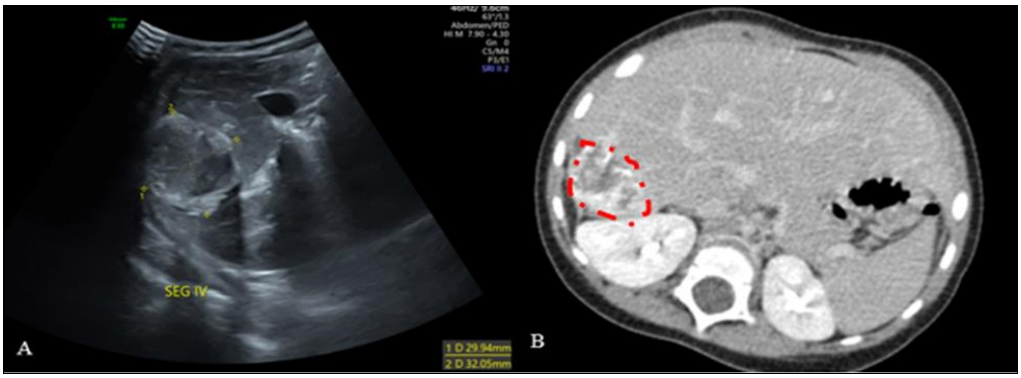

Fig 3: Follow-up ultrasound (A) and CT (B) performed after one year of treatment revealed a significant reduction in the size of the necrotic hepatic mass estimated at 91% with the lesion now confined to hepatic segments VI and VII.

Second case

A 1 month male neonate was initially admitted with a presumptive diagnosis of neuroblastoma. He was born at term, and a prenatal ultrasound was performed and had revealed an abdominal mass suggestive of a cystic lymphangioma. APGARS were 4, 6, and 8. Clinical examination at birth revealed a 3/10 respiratory distress, a distended abdomen, and meconium passing.

Upon admission, the neonate was eupneic, with a blood pressure in all limbs at about 70/50 mmHg. The neonate also had a good suckle reflex, a slight hypotonicity, and stage 3 jaundice. The abdomen was distended with no signs of inflammation; no mass in lumbar regions; no hepatosplenomegaly, with a negative screening for malformation.

Laboratory findings revealed thrombocytopenia with a count as low as 36,000/mm³, as well as anemia with a level of 9 g/dL. Other examinations included a level on total bilirubin at 178, direct bilirubin at 7, calcium at 90, creatinine at 6, as well as a level of 0.18 on urea. The TORCH serology was negative, while the urine test was positive. Serum AFP and beta-HCG levels were within normal range.

Admission abdominopelvic ultrasound (Figure 4) shows a massive hepatic mass in segments VIII, VII, IVa, V, and I. This mass displayed an exophytic component bulging inferiorly from the liver. It was roughly oval shape with lobulated borders and heterogenous echotexture, containing vascular lakes. The mass on ultrasound measured 85 x 73 x 101 mm. The remainder of the hepatic parenchyma was homogenous with regular borders with no other focal lesions.

CT (Figure 5) shows a massive intraperitoneal tumor mass which arose from segments IV, V, VI, VII, and VIII of the liver. It has an oval shape with lobulated borders and heterogeneously enhanced on contrast administration. It featured a central zone of hypodensity consistent with extensive necrosis with serpentine vessels in its periphery, and measuring approximately 80 x 61 x 100 mm.

Treatment with propranolol 0.5 mg/kg 12 hourly was initiated for a week, followed by a dose increment to 1 mg/kg 12 hourly for a week, with a final increment to 1.5 mg/kg 12 hourly. The patient also took a quarter tablet of Avlocardyl 40 mg in 5 cc mineral water.

The patient remains under regular ultrasound surveillance, which has shown a partial regression of the mass following treatment.

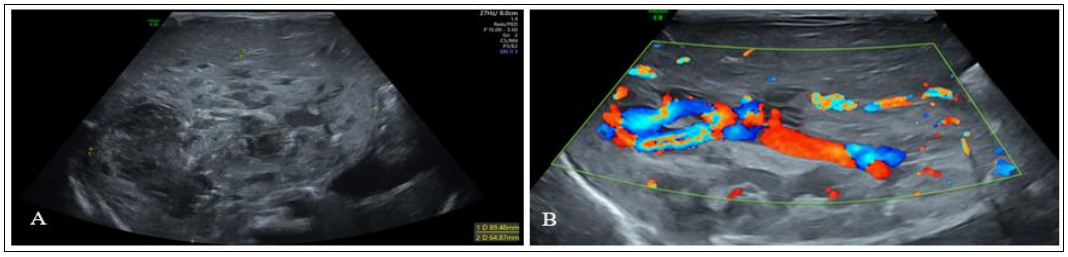

Fig 4: A massive hepatic mass in segments VIII, VII, IVa, V, and I, with an oval shape (A) and lobulated borders. It exhibited a heterogeneous echotexture, containing vascular lakes that were highlighted on color Doppler imaging (B).

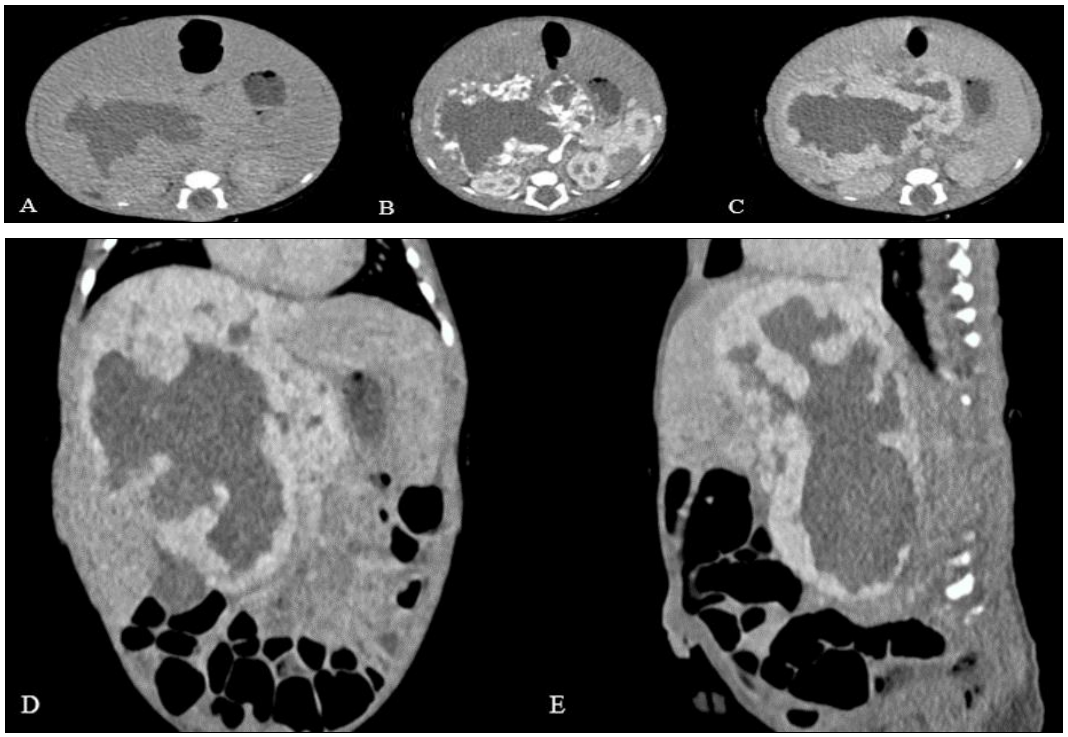

Fig 5: Axial (A,B,C) coronal (D), and sagittal (E) CT sections before and after administration of contrast demonstrated a massive intraperitoneal tumor mass which arose from segments IV, V, VI, VII, and VIII of the liver. The mass was oval in shape with a lobulated border and heterogeneously enhanced on contrast administration. It featured a central zone of hypodensity consistent with extensive necrosis with serpentine vessels in its periphery.

Discussion

Hepatic hemangiomas are the most common infantile benign liver tumors. Although most are asymptomatic in their course, it is not possible in a given case to predict which lesions will be quiet versus which will progress to a cause of symptomatic illness [1, 2, 3]. In addition, whereas many hepatic hemangiomas have a classical pattern on cross-sectional studies, a minority have atypical appearances that if carefully assessed can be identified as a hemangioma [4, 5].

Clinical Presentation

IHHs are very heterogenous in terms of presentation. In the past, these lesions have been reported in a 3:1 (F/M) ratio, though some more contemporary series report ratios as high as 4:3. Referral bias is probably the explanation because those with more serious or atypical presentations are treated in specialized tertiary care units [6, 7]. The vast majority of IHH are incidentally detected and do not require treatment because of their benign clinical course, some patients present with complications that necessitate intervention.

One very suggestive clinicopathologic correlation is that in those with extensive hemangiomatous liver involvement, some will have hypothyroidism (which is the case in our first clinical case report).

Imaging Features

Imaging is crucial in both IHH diagnosis and treatment planning. The cross-sectional imaging techniques of CT, MRI, and US are required to measure lesion dimensions, morphology, blood supply, and relation with adjacent structures.

Multifocal Hemangiomas

Multifocal hepatic hemangiomas are spherical lesions with homogenous signal intensity on MRI displaying decreased signal on T1-weighted images and markedly increased signal on T2-weighted images. The lesions are generally observed with vascular flow voiding around or between the nodules, reflecting high-flow dynamics with dilated hepatic arteries and veins. Following administration with a contrast agent, these lesions enhance homogeneously or in a classical centripetal pattern [8, 9].

Focal Hemangiomas

Focal hemangiomas are more heterogenous in appearance. Centripetal enhancement is common, with a high frequency in many lesions having non-enhancing central regions due to necrosis. High-flow variants can demonstrate direct arteriovenous or portovenous shunts, with a central varix and aberrant draining hepatic veins.

Ultrasound and Doppler Evaluation

Ultrasonography remains the modality of preference in the investigation of presumed IHH. The sonographic features are variable with hypoechoic to hyperechoic lesions that are often seen with a background of aberrantly flowing as well as shunted vessels. The use of Doppler imaging enhances diagnostic accuracy by demonstrating the presence of dilated vessels and abnormal flow patterns however, in cases of diffuse liver involvement, individual nodules may be difficult to delineate [10].

Mri and Advanced Techniques

MRI, particularly in combination with dynamic contrast-enhanced and gradient echo sequences, is considered the most sensitive and the most specific modality in characterizing hepatic hemangiomas. Its high tissue contrast with its ability to examine vascular architecture makes it critical in distinguishing common hemangiomas from intrahepatic mass lesions. Recent technical developments in multidetector CT angiography as well as advanced MR angiographic techniques have also improved assessment in detecting shunts. However, angiography is generally reserved in symptomatic shunts in whom endovascular treatment is planned [4, 5].

Therapeutic Challenges and Management Strategies

Treatment of IHH is a tough choice that balances the relatively innocuous nature of most lesions with a risk of fatal complications. There is a range of treatments available, each with its own risk profile.

Medical Management

Long-standing in medical management of IHH are steroids, with interferon and vincristine as alternative treatments in case steroids are not effective or are contraindicated. The therapies are not risk free. The growth retardation and immunosuppression are some side effects of steroids, while interferon and vincristine have also been associated with severe adverse outcomes in terms of cardiomyopathy, and spastic diplegia [11].

Interventional Methods and Surgeries

Imaging findings, particularly the identification of vascular shunts, play a crucial role in establishing whether interventional or surgical treatment is warranted. Evident arteriovenous, arterioportal, or portovenous shunts in patients are far more likely to be treated with embolization or surgical resection as compared with those with no shunts [9]. In extensive liver involvement in which pharmacotherapy is not effective, patients can develop a complication in terms of abdominal compartment syndrome or hemorrhagic shock. Such nonresponsive patients can ultimately be treated with a transplant of the liver [12].

Conclusion

Overall, whereas the majority of infantile hepatic hemangiomas are not symptomatic, a subset evolves into severe clinical complications that require early aggressive management. The vast range in clinic presentation from incidental detection to life-threatening illness in high-output cardiac failure and hypothyroidism requires a tailored, multidisciplinary approach. In this context, imaging is essential with MRI as modality of choice, supported by ultrasound and CT in initial investigation and follow-up.

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- **Data access statement:** All data supporting the findings of this study are included in the article. No additional datasets were generated or analyzed.

- **Ethics statement:** This case report was conducted in accordance with ethical standards. Informed consent was obtained from the patient for the publication of this case and accompanying images.
- **Competing interests:** The authors declare that they have no competing interests.

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