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Case report

Hepatic mesenchymal hamartoma: The role of radiology in diagnosis and management ☆,☆☆

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ABSTRACT

Hepatic mesenchymal hamartoma is an uncommon tumor composed of myxoid mesenchymal tissue with fluid accumulation. Here, we report a case of hepatic mesenchymal hamartoma in a 3-year-old patient who was referred to the hospital with abdominal pain and a slight fever. An abdominal ultrasound suggested a liver abscess; however, computed tomography revealed a mass with both cystic and solid components. Liver biopsy confirmed hepatic mesenchymal hamartoma, and this patient underwent the complete removal of this tumor.

Introduction

Hepatic mesenchymal hamartoma (HMH) is a rare, benign tumor that occurs in pediatric patients [1], often affecting children younger than 2 years [2]. The etiology of HMH is unclear, although HMH has been associated with polycystic kidney, congenital liver fibrosis, biliary atresia, congenital heart diseases, and biliary hamartoma [3]. HMH may present as a large, benign, cystic mass, as a solid mass, or as a mass with mixed cystic and solid components [1]. The optimal therapy modality for HMH is complete resection [3]. Few descriptions of LMH

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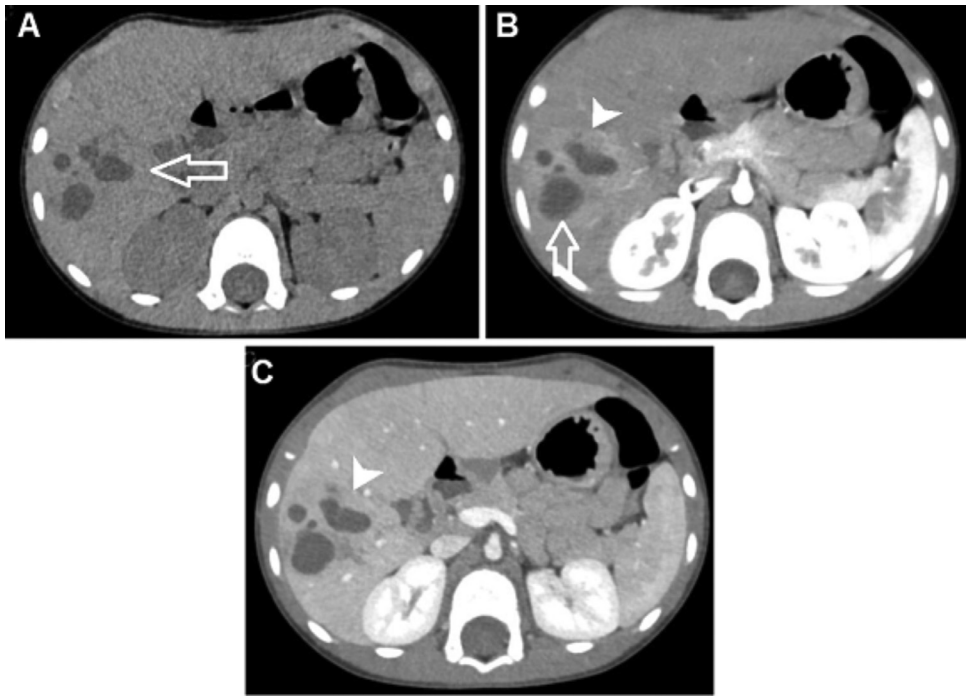

Fig. 1 – Abdomen computed tomography (CT) scans. (A) Noncontrast CT scans showed a lesion with a mixture of cystic and solid components in hepatic segment VI (arrow). (B) On the arterial phase, the cystic components did not enhance (arrow), but the solid components showed hypervascularity (arrowhead). (C) On the venous phase, the sold components were less enhancing than the normal liver parenchyma (arrowhead)

have been identified in the literature. We present this case to provide additional and useful information regarding the identification and treatment of this disease.

Case report

A 3-year-old boy presented with chronic and vague abdominal pain for 1 month, with no history of jaundice, fever, or weight loss. Clinical examination detected no significant abnormalities, with the exception of a slightly elevated temperature (37.8°C). Abdominal ultrasound showed a heterogeneous lesion with cystic components in hepatic segment VI. Abdominal computed tomography (CT) scans revealed a complex solid–cystic lesion with well-defined borders in the hepatic segment, with the size mm (Fig. 1). The cystic components were not enhancing, and the solid components were more enhancing than the normal hepatic parenchyma in the arterial phase, whereas, in the venous phase, the solid components were less enhancing than the normal hepatic parenchyma (Fig. 1). Laboratory studies revealed serum alanine transaminase levels: 20 IU/L (normal range 10–40 IU/L); aspartate transaminase: 30 IU/L (normal range 0–40 IU/L); total bilirubin: 0.15 mg/dL (normal range 0.1–1.4 mg/dL); and alpha-fetoprotein: 4.5 IU/mL (normal range 0–6 IU/mL). The complete blood count was normal. Because the CT scan suggested a liver mass, a liver biopsy was performed, and the histological results confirmed HMH. Complete tumoral resection was performed, and 1 year after surgery, the patient showed no signs of recurrence.

Discussion

HMH often occurs in pediatric patients and accounts for 8% of all pediatric tumors [2], with a slight male predominance [4]. The symptoms of this disease are variable, and patients may be asymptomatic or present with abdominal distension, upper abdominal palpable mass, abdominal pain, anorexia, vomiting, and poor weight gain [3,5]. Other presentations include cardiac failure, obstructive jaundice, and symptoms due to spontaneous abscess formation [3]. A large tumor can rupture, leading to bleeding, which can be life-threatening [3].

Serum alpha-fetoprotein levels are typically normal but can also be elevated, and elevated levels return to normal after complete tumor resection [3,5,6]. Liver function tests are also generally normal [5].

Approximately 75% of HMH cases develop in the right lobe of the liver [7]. On imaging, the appearance of HMH may be variable, appearing as a multiseptated cystic tumor, a mixed

cystic and solid tumor, or solid tumor, depending on the HMH components [1]. These tumors are often well-circumscribed with varying sizes of up to 30 cm [6]. Hemorrhage and calcification are uncommon [8]. On CT scans or magnetic resonance imaging with contrast agents, the septa appear enhanced, and the solid components are often heterogeneously enhanced, which persists on the delayed phase [1,3,6]. Some differential diagnoses for HMH include hemangioma, hepatoblastoma, undifferentiated embryonal sarcoma, and parasitic infections [9]. HMH can be difficult to differentiate from other tumors due to the nonspecific imaging features, and biopsy is necessary in these cases.

The diagnosis of HMH is based on the histological examination. On microscopic examination, the tumor consists of mesenchymal and epithelial components [3]. The stroma contains fibroblasts; myxoid stroma, which can be collagenous or hyalinized; bile ducts; blood and lymphatic vessels; and islands of hepatocytes [3,10]. The cysts are lined with epithelium, and the bile ducts and hepatocytes tend to be located in the periphery of the lesion [3]. Surgical resection with a negative margin is the standard treatment for HMH tumors, and incomplete removal can result in recurrence [2].

In this case, the patient presented with symptoms quite similar to infection, associated with a heterogeneous, echoic lesion on ultrasound, which was initially diagnosed as a liver abscess. However, on CT scans, the lesion did not present as a typical abscess, featuring well-defined borders with cystic and solid components and the solid components enhanced homogeneously. The patient underwent a biopsy, which resulted in a diagnosis of HMH, and the tumor was completely removed.

Conclusion

In conclusion, HMH in children is uncommon and can present with various features on imaging. However, CT scans may suggest a tumor with features that differ from other benign lesions, as in this case. This disease can be completely cured using surgical excision as the primary therapeutic modality.

Ethical statement

Appropriate written informed consent was obtained for the publication of this case report and accompanying images.

Author contributions

Tran PN, Truong QD, and Nguyen MD contributed to this article as co-first authors. All authors have read the manuscript and agree to the contents.

REFERENCES

[1] Kim SH, Kim WS, Cheon JE, Yoon HK, Kang GH, Kim IO, et al. Radiological spectrum of hepatic mesenchymal hamartoma in children. Korean J Radiol 2007;8(6):498. doi:10.3348/kjr.2007.8.6.498.
[2] Cahís Vela N, Loverdos Eseverri I, Beltrán Salazar VP, Brun Lozano N, Ferreres Piñas JC, Hernández CJR. Liver mesenchymal hamartoma. J Pediatr Surg Case Rep 2021;68:101839. doi:10.1016/j.epsc.2021.101839.
[3] Stringer MD, Alizai NK. Mesenchymal hamartoma of the liver: a systematic review. J Pediatr Surg 2005;40(11):1681–90. doi:10.1016/j.jpedsurg.2005.07.052.
[4] Anil G, Fortier M, Low Y. Cystic hepatic mesenchymal hamartoma: the role of radiology in diagnosis and perioperative management. Br J Radiol 2011;84(1001):e91–4. doi:10.1259/bjr/41579091.
[5] Hassan MA. Giant solid mesenchymal hamartoma of the liver in a neonate: case report. Ann Pediatr Surg 2020;16(1):29. doi:10.1186/s43159-020-00040-0.
[6] Rosado E, Cabral P, Campo M, Tavares A. Mesenchymal hamartoma of the liver - a case report and literature review. J Radiol Case Rep 2013;7(5):35–43. doi:10.3941/jrcr.v7i5.1334.
[7] Koganti SB, Thumma VM, Nagari B. Mesenchymal hamartoma of the liver: complete excision always necessary. Case Rep Surg 2017;2017:1–4. doi:10.1155/2017/8314102.
[8] Armanian AM, Nazem M. Mesenchymal hamartoma of liver in a neonate. J Neonatal Biol 2016;05(02). doi:10.4172/2167-0897.1000223.
[9] Gray SC, Pienaar JA, Sofianos Z, Varghese J, Warnich I. Hepatic mesenchymal hamartoma: an uncommon but important paediatric diagnosis. SA J Radiol 2020;24(1):1891. doi:10.4102/sajr.v24i1.1891.
[10] Chang HJ, SY Jin, Park C, Park YN, Jang JJ, Park CK, et al. Mesenchymal hamartomas of the liver: comparison of clinicopathologic features between cystic and solid forms. J Korean Med Sci 2006;21(1):63–8. doi:10.3346/jkms.2006.21.1.63.