Case Report

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Giant hepatic capillary hemangioma, a diagnostic dilemma: case report

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ABSTRACT

Hepatic Hemangiomas (HH) are benign tumors of the liver consisting of a blood-filled cavity supplied by the hepatic artery. Most of the HH are asymptomatic and are discovered incidentally during radiological imaging of various unrelated pathology. Typical capillary hemangiomas range from a few millimeters to three centimeters and are unlikely to generate any future symptomatology. However, giant liver hemangiomas (more than 10 cm) are a very rare entity and might become symptomatic, hence requiring prompt intervention. We are reporting this case to make the readers aware of the pitfalls and radiological uncertainties while interpreting vascular lesions of the liver. We reported a case of a 21-year-old female with complaint of awareness of a mass over the right upper abdomen since a period of one year which has been gradually increasing in size. On pre-operative Contrast-Enhanced Computed Tomography, features suggestive of Fibrolamellar variant of Hepatocellular Carcinoma was found which was substantiated by biochemical investigations. However, post-operative histopathological examination revealed a capillary hemangioma.

Keywords: Giant capillary hemangioma of liver, Fibrolamellar hepatocellular carcinoma, Benign hepatic tumor, Hepatic lobectomy

INTRODUCTION

Hepatic vascular tumors include cavernous hemangioma, infantile hemangioendothelioma, epithelioid hemangioendothelioma, angiosarcoma and anastomosing hemangiomas. Most hemangiomas of livers are cavernous hemangiomas which are distinct from hepatocellular carcinoma on radiological imaging. 1-4

Hepatic hemangiomas, being a rare pathological entity with often unclear clinical features, bring about a diagnostic confusion with hepatocellular carcinoma or cavernous hemangioma. We describe a case of giant capillary hemangioma of the liver. This is a unique case compared to those reported in earlier literature, courtesy to the associated features and discuss issues concerning diagnosis and management.

CASE REPORT

A 21-year-old female patient presented with a complaint of insidious onset of awareness of a painless abdominal mass on the right upper abdomen, which had been progressively increasing in size over the past year. There was no history of jaundice, hematemesis, melena or altered bowel habits. There was no history of fever, loss of appetite or loss of weight. Family history and past medical history were inconclusive. On per abdominal examination, a non-tender, hard, intra-abdominal, intra-peritoneal ovoid mass of approximately 13×9 cm was felt in the right upper quadrant which was around 12 cm below the right costal margin. It was not possible to get over the swelling. A differential diagnosis of benign hepatic tumor, fibrolamellar hepatocellular carcinoma, abdominal parietal wall tumor and mucocele of the gall bladder was considered. Ultrasonographic (USG) images showed a

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heterogeneous space-occupying lesion in the liver with a size of 10×8.8 cm and unclear blood flow, both in and out of the tumor on color doppler images. There was no evidence of a septum or a lateral shadow. Upper gastrointestinal endoscopy showed normal study.

A Contrast-enhanced Computed Tomography was done after the initial Ultrasound, which showed the liverenlarged in size with a lobulated outline and heterogeneous contrast attenuation. A large $24 \times 12 \times 8$ cm, isodense to hypodense mass with lobulated outline mass of soft tissue density and predominant exophytic component was seen to arise from the right lobe, involving almost whole of the right lobe. Focal non-enhancing scar seen in the mass. No IHBRD. The hepatic artery was dilated and tortuous with the right branch supplying the mass. IVC was compressed, likely to be fibrolamellar HCC (Figure:1-3).



Figure 1: CECT showing focal non-enhancing scar in the mass.

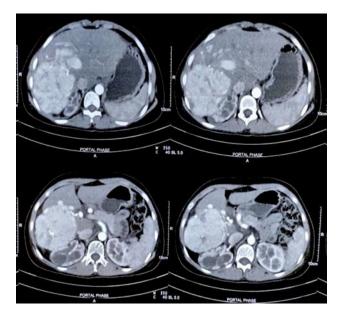


Figure 2: A large 24×12×8 cm, isodense to hypodense mass with lobulated outline, mass of soft tissue density and predominant exophytic component was seen to arise from the right lobe.



Figure 3: Lobulated outline of the mass seen.

Biochemical investigation of tumor markers [Alphafetoprotein (AFP)], CA 125, CA 19-9, Carcinoembryonic antigen (CEA), Beta Human chorionic gonadotropi (β-hCG) were within normal limits. Serial pre-operative laboratory investigations of Liver Function Tests were within normal limits.

Given the patient's complaint of an increasing size of the mass and the CECT suggesting a malignant etiology, right hepatic lobectomy with caudate lobe resection was advised to the patient. After informed consent, the patient was taken up for the surgical procedure under general anesthesia. Intraoperatively, a huge pale yellow solid telangiectatic mass (Figure:4) of approximately 15×10 cm size was seen to arise from the right lobe of the liver extending inferiorly and the gall bladder was pushed medially, visible pulsatile veins were found over the surface of the lesion. An enlarged common hepatic artery and all its branches were seen. The post-operative period was uneventful. The surgically resected lesion was sent for histopathological examination which revealed the definitive diagnosis of capillary hemangiomas of the liver.



Figure 4: Gross specimen of huge pale yellow solid telangiectatic mass.

DISCUSSION

Hepatic Hemangiomas are characterized by capillary or cavernous growth resulting from proliferative blood vessels.1 In the newborn, capillary hemangiomas of the liver were reported to be a symptom of diffused neonatal hemangiomatosis or Kassabach-Merritt syndrome.⁵ Overall, adult hepatic capillary hemangioma is a rare entity, and to our knowledge, only a few have been reported. Most hepatic hemangiomas are asymptomatic or if symptomatic, they are nonspecific, common to many other diseases especially of digestive origin (pain in right upper hemi-abdomen, decreased appetite, nausea, vomiting, post-prandial bloating).⁵⁻⁷ Although physical examination very rarely detects a palpable mass, in our case, a distinct mass was palpable because of its large size.⁷ Pre-operative Contrast-Enhanced Computed Tomography showed features suggestive of fibrolamellar HCC and normal levels of Alpha-Fetoprotein. This also pointed to the same diagnosis, which warranted a prompt surgical intervention because of its malignant nature. However, post-operatively gross pathological examination of the resected specimen was doubtful to be typical HCC or typical hemangioma. Histopathological examination revealed confirming it to be capillary hemangioma (Figure 5).

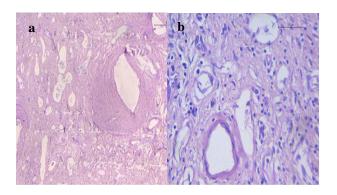


Figure 5 (a and b): H and E 40X and 100X image of the biopsy specimen showing hepatic capillary hemangioma.

The radiological features are inconclusive and might often lead to misinterpretation of definitive diagnosis; hence histopathological examination stays as the gold standard of investigation. Despite this, percutaneous biopsies of doubtful hepatic lesions should be done cautiously on a case to case basis weighing risk: benefit ratio as vascular tumors which don't have characteristic radiological features, can be missed and post-biopsy have a greater risk of hemorrhage (especially in large sub-capsular lesions) even causing mortality.^{8,9} Moreover, the diagnostic yield is not as high as expected: Terriff BA et al. described it in with 36 patients where diagnostic histopathological material was obtained in only 21 of them.¹⁰ Hence biopsy is reserved for extremely atypical lesions with equivocal features on imaging. Keeping the definitive diagnosis of capillary hemangioma in mind, a PubMed search was carried out using the words, "capillary

hemangioma", "liver", "central scar" was carried out which yielded no results. So, we are reporting the diagnostic challenges we faced while working up the case as a central scar in a liver lesion is mostly seen in focal nodular hyperplasia and the fibrolamellar variant of HCC. Literature review revealed that management by supervision through imaging methods, every 6 months or annually, to assess the scale of development over time is sufficient.⁷ Long-term follow-up is warranted in patients who have pain unresponsive to analgesics or receiving estrogen therapy and is mandatory for those with giant hepatic hemangioma. There is no known pharmacological therapy for reducing the size, although anti-angiogenic therapy with Bevacizumab was considered without confirmation.⁷ Surgical intervention reserved for cases with a rapid increase in size or risk of intra-tumoural thrombosis includes segmental resections, lobectomy, or enucleation by open surgery or laparoscopy. 11 For all techniques, post-operative morbidity is minimal.

Other intervention includes angiographic embolization of the portal vein, selective irradiation of the liver, radiofrequency ablation and surgical trans hepatic ligation of the major filling vessels. 12-18 Orthotopic liver transplantation is indicated for large lesion involving both lobes. 2

CONCLUSION

This case has an unusual presentation. Although most adult hepatic hemangiomas are non-progressive and do not require treatment, there are a small number of cases with rapid volumetric growth or complications that prompt appropriate therapy.⁷ To our knowledge, very few cases of adult hepatic Hemangiomas have been reported.^{19,20} We wanted to report this case to make the readers aware of the radiological pitfalls in diagnosing such atypical lesions.

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