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## Case report

# A case of diffuse hepatic hemangiomatosis coexistent with giant hemangioma: case report and literature review <sup>☆</sup>

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## ABSTRACT

We report a case of hepatic hemangiomatosis coexistent with a giant hepatic hemangioma diagnosed in the context of the study for SARS-CoV-2 infection. Computed tomography showed irregular contours of the left hepatic lobe with a lesion that compromised the whole lobe, with peripheral uptake and a centripetal tendency in the late phase, compatible with bulky cavernous hemangioma, as well as another lesion with the same characteristics in segment VI. Magnetic resonance imaging of the abdomen showed multiple hepatic hemangiomas observed in both lobes involving all segments, some of them smaller than 1 cm, which were hyperintense in the T2-weighted sequences and showed progressive contrast enhancement. This case illustrates the incidental diagnosis of this condition during the study for another pathology, the radiological features that are important to differentiate from other tumoral findings, and the possible management strategies to follow.

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## Introduction

The widespread use of imaging modalities and the continuous improvement in their sensitivity have led to an increasing number of incidental findings of focal liver masses, either solitary or multiple. Most of these so-called incidentalomas are benign and are discovered in healthy, asymptomatic patients [1]. Hepatic hemangioma is the most prevalent type of benign liver tumor with an incidence reaching 20% in the overall population, detected by upper ab-

dominal ultrasound [2]. Hemangiomas occur more commonly in the liver than in any other internal organ, although symptomatic massive hepatic hemangiomas are rare [3].

Diffuse hepatic hemangiomatosis is a rare benign condition characterized by diffuse replacement of liver parenchyma with hemangiomatous lesions. It can occur in all age groups, but it is most often detected in neonates whose entire liver is usually involved. The etiology and clinical course are not completely understood because of the rarity of the condition. It has been reported in patients with hereditary hemorrhagic telang-

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iectasia or associated with hemangiomas of the skin and involvement of at least two visceral organs [4].

We report one case of a 63-year-old man, diagnosed with diffuse hepatic hemangiomas of the right hepatic lobe co-existent with left giant hepatic hemangioma, in the context of a study for SARS-CoV-2 infection.

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## Case report

A 63-year-old man with a past medical history of paranoid schizophrenia, urethral stricture, and duodenal ulcer, with no known drug allergies, consulted our emergency room due to fever, cough and dyspnea. He denied any alcohol or illicit drug use. His medications included amisulpride, venlafaxine, temazepam, levosulpiride, omeprazole, levomepromazine and diazepam.

The physical exam presented tachypnea and abundant bilateral crackles, with pulse oximetry saturation of 78% on room air, and the chest X-Ray showed bilateral white-cottony infiltrates. As the patient presented acute hypoxemic respiratory failure secondary to bilateral pneumonia, with suspicion of COVID-19, he was admitted to the Intensive Care Unit (ICU).

The result of a real-time reverse transcriptase–polymerase chain reaction (RT-PCR) for SARS-CoV-2 was positive. During his stay in the ICU, the patient required orotracheal intubation and mechanical ventilation, posterior tracheostomy, and prone position. He was treated with hydroxychloroquine, ceftriaxone, and azithromycin. Progressive weaning was conducted until decannulation without incident. Chest CT (CCT) showed alteration of the lung radiological pattern with bilateral ground-glass opacities, consistent with the viral infection [5] (Fig. 1).

Among the complications, he presented symptoms of abdominal bloating and constipation. Physical examination showed hepatomegaly in the right hypochondrium, with no pain associated. An abdominal CT was performed showing feces and fecaloma, oriented as a paralytic ileus in the context of the treatment and the immobilization. After laxative treatment, the patient presented bowel movements. On the other hand, the same CT showed irregular contours of the LHL with a neoplastic process that compromised the entire LHL, showing peripheral uptake with a centripetal tendency in the late phase, compatible with bulky cavernous hemangioma, as well as another lesion with the same characteristics in segment VI of the right hepatic lobe of 2.5 cm (Fig. 2).

The patient was transferred to the regular hospital ward. The laboratory tests including ALT, AST, ALP, GGT, bilirubin (total and direct), albumin, amylase, LDH, and prothrombin time were normal. Alpha-fetoprotein was 4.05 ng/mL. Serum tumor markers were all within the normal range. Hepatitis B and C virus serum markers were negative.

The study was completed using doppler ultrasonography and MRI. The first one showed the liver with heterogeneous echo structure and the presence of a rounded solid-liquid area of approximately 70 mm that compromised the left lobe, suggestive of probably cavernous angioma, and another area of 20 mm in segment V. No solid-type focal lesions were observed. The Portal vein preserved its caliber with hepatopetal flow of

15 cm/sec. The Suprahepatic vein was leaky. No signs of umbilical vein recanalization or gastric vein dilation were observed.

The MRI of the abdomen showed the liver enlarged, with signs of hepatomegaly: a 21 cm right hepatic lobe in the cranio-caudal axis and a 14 cm left hepatic lobe; showing no signs of liver iron, fat deposition or signs of chronic liver disease. Multiple hepatic hemangiomas were observed in both lobes involving all segments, some of them smaller than 1 cm, which were hyperintense in the T2-weighted sequences, showing progressive contrast enhancement. A giant hemangioma involving segments II, III, IV-A, IV-B, VIII, and V was observed. This hemangioma had a maximum transverse diameter of 21 cm and an anteroposterior diameter of 9 cm, and was markedly hyperintense in T2, corresponding to a progressive centripetal nodular filling. Another hemangioma was also observed between segments VIII and VII, with a maximum diameter of 6 cm, and another hemangioma in segment VI toward the posterior slope, with a maximum diameter of 4 cm; the rest of the hemangiomas measured less than 1 cm (Fig. 3). These characteristics were consistent with the diagnosis of liver hemangiomas [6] and biopsy was not performed due to the absence of chronic liver disease, normal laboratory tests and the evaluation of the imaging of the liver that showed the vascular nature of the lesions.

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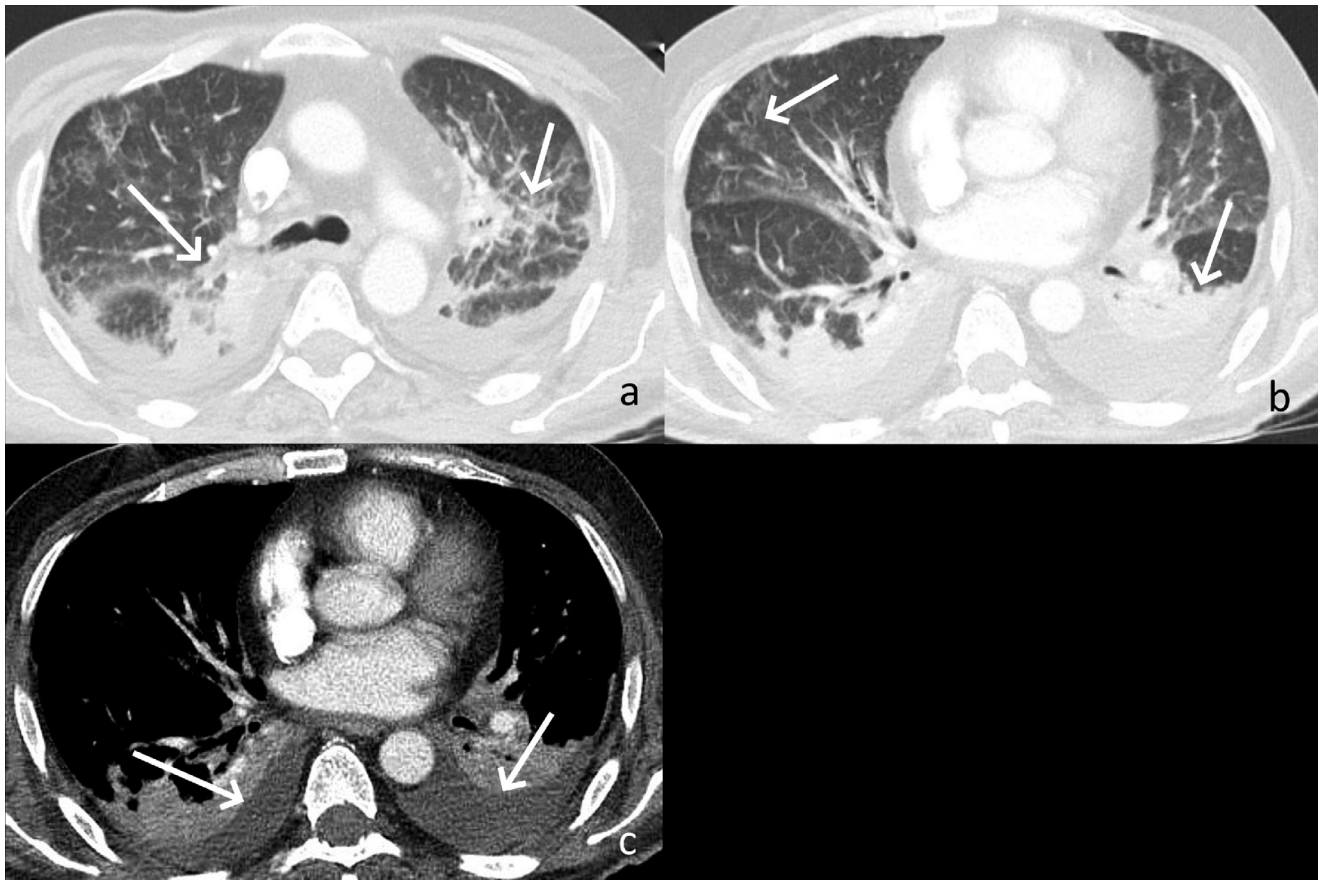
## Discussion

The hepatic hemangioma is the most common of benign liver tumors incidentally detected in most patients. They are often solitary, although multiple lesions may be present in both hepatic lobes in up to 40% of the patients. Hepatic hemangiomas are also known as cavernous hemangiomas. Their size varies from a few millimeters to over 20 cm. Those lesions larger than 5 cm are considered giant hemangiomas [7]. These tumors are generally found by radiological imaging and, of such examinations, nuclear magnetic resonance is the best diagnostic method. Most cases are asymptomatic and must be followed up by periodic radiological examination. Surgical treatment should be restricted to unusual situations, such as spontaneous or traumatic rupture, Kasabach-Merritt syndrome, uncertain diagnosis, and pain [8].

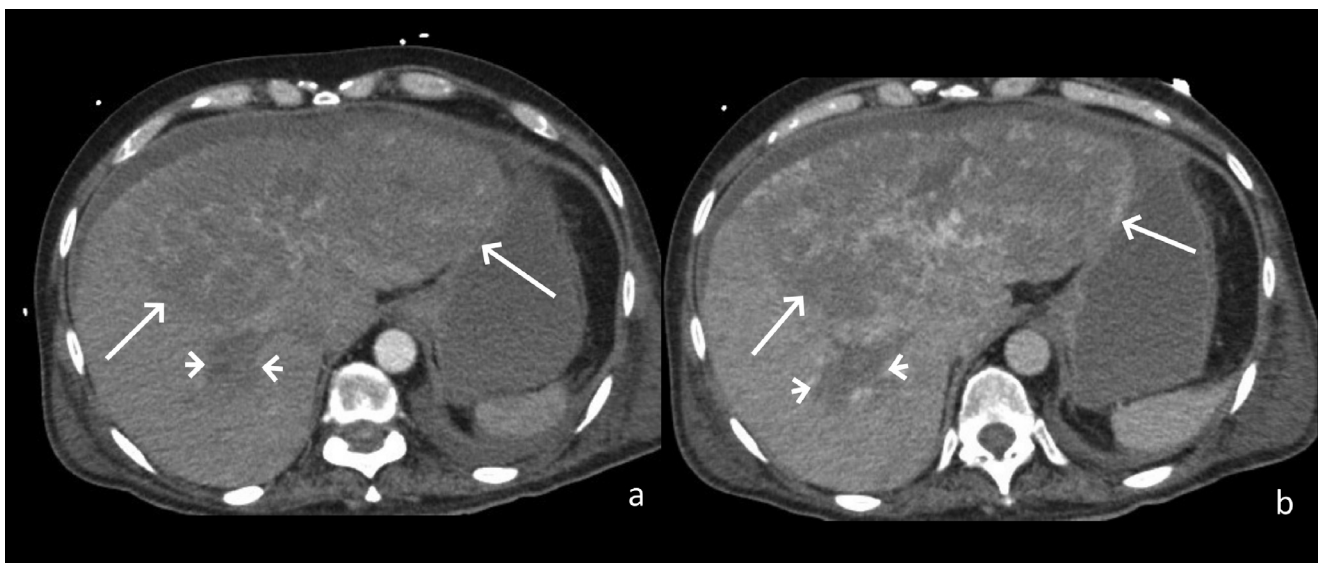
On the other hand, hepatic hemangiomas replacing the entire hepatic parenchyma is extremely rare in adults. The natural history of hepatic hemangiomas and factors that modulate tumor growth are poorly understood because of its rarity [9], while it is often reported in infants associated with Rendu-Osler Weber disease, and may cause serious complications [10].

Isolated liver hemangiomas, without any extrahepatic involvement or association with giant hemangioma, is uncommon in children and even more so in the adult population. In 2018, Liu et al. Little presented two cases of isolated hepatic hemangiomas, and included a review of 15 other cases of isolated hepatic hemangiomas reported in English literature from 1970 up to that moment (July 2018) [6].

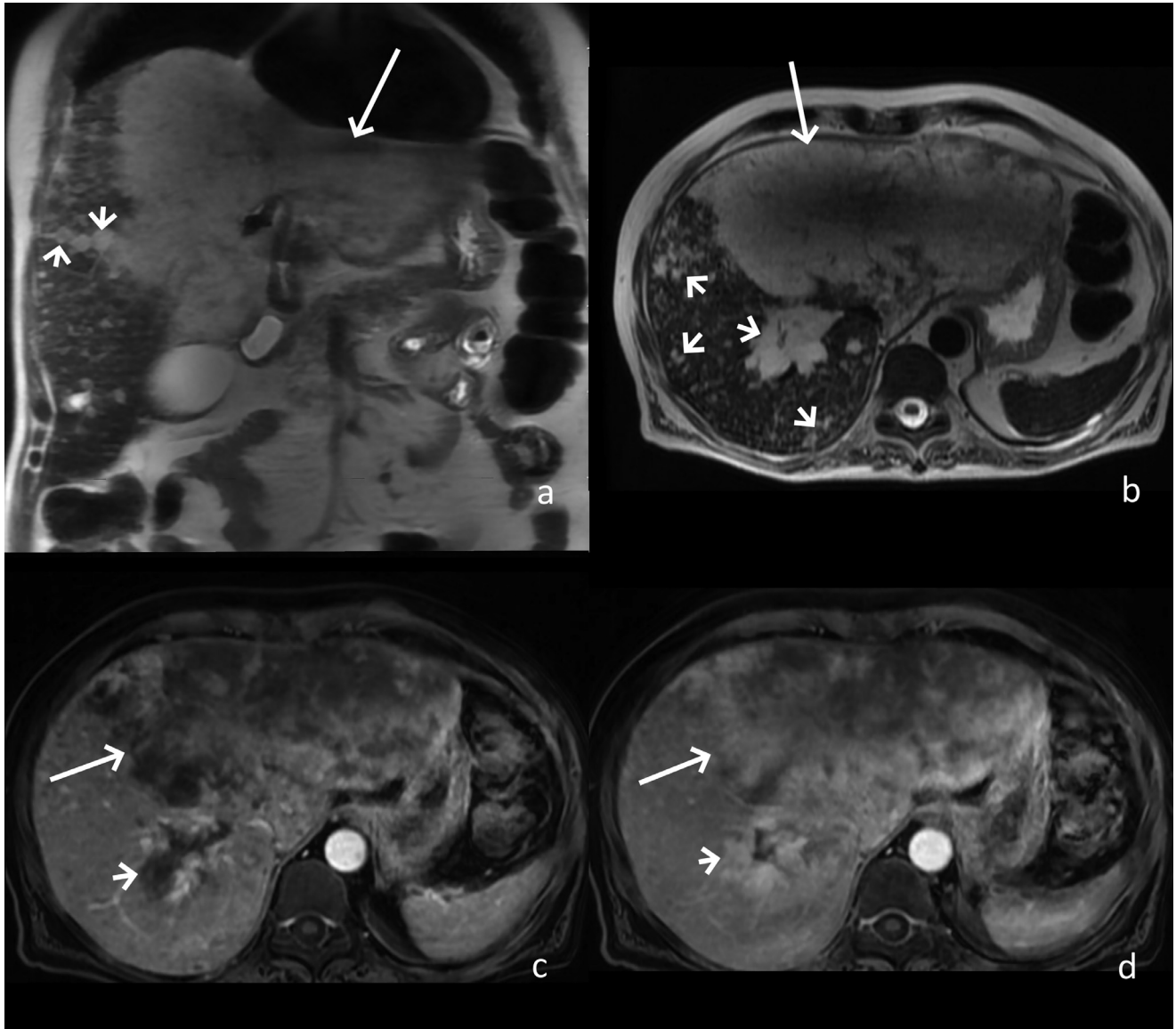
However, hemangiomas seems not uncommon in the liver parenchyma adjacent to a giant cavernous hemangioma (GCH). In 2011, Jhaveri et al., through a retrospective study us-



**Fig. 1** – Contrast-enhanced chest CT. SARS-CoV-2 infection. Multifocal and bilateral ground-glass opacities (arrows) in both upper (A) and lower (B) lobes with a central and peripheral location. Bilateral lower lobe consolidations (C) with bilateral pleural effusion (arrows).



**Fig. 2** – Contrast-enhanced abdominal CT: hepatic hemangiomas with giant hemangioma. Arterial (A) and venous phases (B). A large hypodense hepatic lesion (arrows) with progressive nodular centripetal enhancement in segments II, III, and IV. Other lesion with similar features (arrowheads) in segments VIA-b and VIII.



**Fig. 3 – Abdominal MRI: hepatic hemangiomas with giant hemangioma. Coronal (A) and axial T2 (B) weighted MR images show an enlarged hepatic liver with multiple well defined hyperintense lesions (arrowheads) involving both hepatic lobes. The largest lesion (arrows) involves the left lobe with 21 cm of largest size (giant hemangioma). Another hemangioma between segments VIII and VII with a maximum diameter of 6 cm is also observed. Axial contrast-enhanced T1 fat sat weighted MR images on arterial (C) and venous (D) phases show centripetal progressive nodular enhancement filling of the lesions.**

ing their imaging database, described hepatic hemangiomas in 18 out of 41 patients who were diagnosed with giant cavernous hemangiomas (44%). This hemangiomas involved the liver parenchyma adjacent to the margins of the GCH without identifiable normal liver tissue separating them in the majority: 17/18 [9].

Additional reports of hemangiomas associated with a giant hemangioma have been described after that one [4,11–16].

The management strategy for giant hepatic hemangiomas can differ depending on the presence of associated hemangiomas and the amount and distribution of residual hep-

atic parenchyma [9]. Symptomatic patients with giant hemangiomas and associated extensive hemangiomas can be difficult to enucleate, and surgical candidates have to be carefully selected to avoid surgical complications related to excessive blood loss from the areas of hemangiomas after the enucleation of the GCH [9]. Maeda et al. presented two cases that were both complicated by consumption coagulopathy (Kasabach–Merritt syndrome). The patients were successfully managed surgically, although through living donor liver transplantation in one case and extensive right lobectomy in the other [15]. Parsia et al. presented another case of a patient with a unique syndrome of multiorgan cavernous hemangioma-

sis involving the liver and other organs. The patient suffered from intra-abdominal hemorrhage due to her giant cavernous hepatic hemangioma. The evidence of continued bleeding, in the setting of Kasabach-Merritt Syndrome and worsening abdominal compartment syndrome, prompted MELD exemption listing. The patient underwent emergent liver transplantation without complication [17].

Hepatic arterial embolization has been reported as the second most common form of treatment of symptomatic giant hemangiomas, which provides symptomatic relief, as well as a reduction in hemangioma size [17,18]. Alternative management techniques include irradiation therapy, which doesn't succeed in the disappearance of the tumor, but arrests the growth and relieves the symptoms. In instances where both liver lobes are diffusely involved or the magnitude of the operation contraindicates it, this method could be preferable, however, the relief of the symptoms could be partial [19].

These other methods rarely have been reported to arrest progression or to improve the symptoms in patients with diffuse hemangiomatosis [9].

Because management strategies of patients with GCH depend on the presence of hemangiomatosis, this association must be clear for the physicians involved in the patient's care, and the amount and distribution of residual hepatic parenchyma needs to be assessed with imaging [9].

## Conclusion

We report a case of a 63-year-old-male patient presenting asymptomatic giant hepatic hemangioma with diffuse hepatic hemangiomatosis, accidentally diagnosed during hospital admission secondary to COVID-19. Chest radiological images showed the characteristic findings of SARS-CoV-2 infection: alteration of the radiological pattern of the lung with diffuse bilateral ground-glass opacities. Abdominal radiological images on CT and MRI showed a neoplastic process that compromised the entire LHL, compatible with a bulky cavernous hemangioma, another lesion with the same characteristics in segment VI, and multiple hepatic hemangiomas in both lobes involving all segments, some of them smaller than 1 cm, which were hyperintense in the T2-weighted sequences, showing progressive contrast enhancement. A liver biopsy was not performed. The recommendation for the patient was evolutionary control via liver MRI every 3-6 months, informing the patient and the family of possible complications. We proposed surgical or angiographic management only in case of urgent complication of the giant hemangioma.

## Patient Consent

Written informed consent was obtained from the patient for the publication of this case report, including accompanying images.

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