Letter to the Editor

A growing liver anastomosing hemangioma

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Introduction

Anastomosing hemangioma (AH) is a benign mesenchymal neoplasm that was first recognized in the genitourinary tract in 2009 by Montgomery and Epstein (1). Microscopically, AH is characterized by anastomosing sinusoidal-like vessels accompanied by scattered hobnail endothelial cells reminiscent of splenic parenchyma. Generally speaking, AH is detected incidentally without specific symptoms. No critical diagnostic features are shown on computed tomography (CT) or magnetic resonance imaging (MRI).

AH of the liver is a rare but distinct subtype of hemangioma (2). Imaging features of liver AH have only been described in a few cases (3), and none of these demonstrated any specific imaging perspicacity (4). The MR imaging features of AH vary (5) and include homogeneous hyperintensity on T2-weighted imaging (T2WI), hypointensity on T1-weighted imaging (T1WI), different degrees of hyperintensity on diffusion-weighted imaging (DWI), and persistent enhancement on enhanced sequences.

Case presentation

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

A hepatic nodule was incidentally detected in a 29-year-old man during an ultrasound examination while checking the condition of a hypoechoic lesion 3 years ago. The instant CT examination revealed a low-density nodule with a length of about 21 mm (*Figure 1A*). After 29 months of ultrasound follow-up, the length of the lesion increased to 46 mm (*Figure 1B*). The ultrasound and MRI with the contrast agent of gadobenate dimeglumine after 36 months showed a mass about 53 mm in length (*Figure 1C*). The patient had no medical history of chronic liver disease or cancer. The patient's laboratory test results, including routine blood tests, blood biochemical examinations, and liver function tests, were normal.

An enhanced and prolonged MRI examination of the liver was performed to confirm a diagnosis. MRI demonstrated a well-demarcated mass sized 53 mm × 45 mm × 41 mm in the right liver lobe that showed homogeneous hypointensity on T1WI (*Figure 2A*), hyperintensity on T2WI and DWI, and early significant enhancement (*Figure 2B*). A central scar in the lesion with a 2-minute delayed enhancement mimicked the manifestation of focal nodular hyperplasia (FNH) (*Figure 2C*). There were no obvious abnormalities in the remaining liver. All figures and tables in the article are original.

Discussion

Considering the imaging characteristics of the central scar

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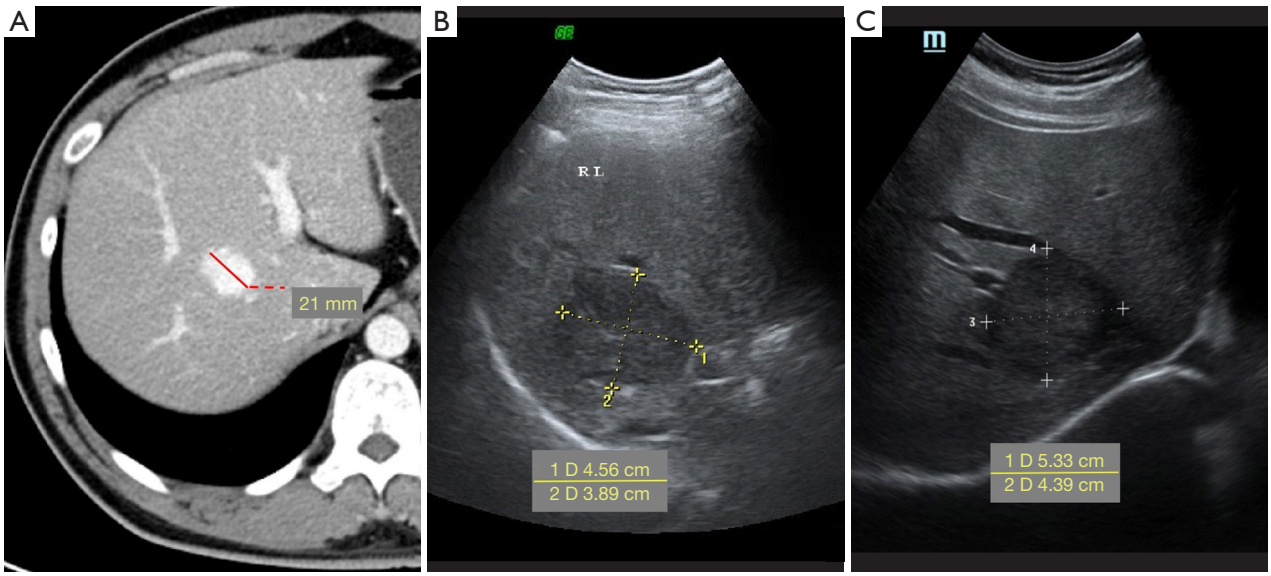

Figure 1 The CT and ultrasound images of AH. (A) The lesion in the liver was first discovered 36 months earlier and the lesion was 21 mm long in the CT image. Ultrasonography showed the lesion had grown to 46 mm after 29 months (B) and to 53 mm after 36 months (C). CT, computed tomography; AH, anastomosing hemangioma.

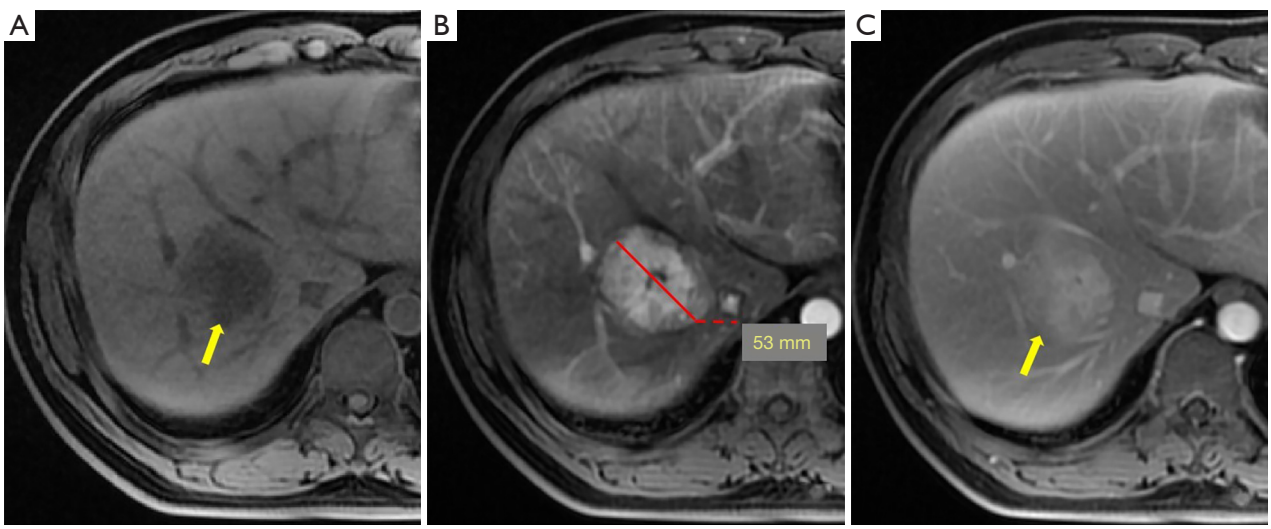

Figure 2 The MRI images of AH. (A) The MRI demonstrated a well-demarcated mass with homogeneous hypointensity on T1WI and (B) early significant enhancement accompanied by a hypointense central scar. (C) The central scar showed delayed enhancement after 2 minutes. The yellow arrow indicates the lesion, and the red line represents the length of the lesion. MRI, magnetic resonance imaging; AH, anastomosing hemangioma; T1WI, T1-weighted images.

with delayed enhancement, the liver mass was preoperatively diagnosed as FNH. Given the rapid growth of the lesion during the 36-month follow-up and after multidisciplinary consultation between radiologists, hepatologists, and hepatobiliary surgeons, it was decided to proceed with operation, after which the lesion was pathologically confirmed as liver AH. This is the first reported case of an intrahepatic AH with MRI features similar to FNH. In this case, the intrahepatic AH continued to grow over 3 years of surveillance. Previous studies mostly focused on the

Table 1 A summary of the previous reports of liver AH

References	Year	Cases (n)	Age (year, range)	Gender (female/male)	Location (left/right lobe)	Size (range, cm)	Conclusions
Lin *et al*. (3)	2013	4	48–71	3/1	3/1	2.0–6.0	AH is a rare distinctive vascular neoplasm that displays overlapping features with well-differentiated angiosarcoma
Merritt *et al*. (2)	2019	1	56	Male	Right lobe	3.4	AH can demonstrate enhancement characteristics similar to primary and metastatic liver lesions
Gonzalez *et al*. (10)	2019	1	80	Female	Left lobe	2.0	AH is a recently described and rare variant of capillary hemangioma
Lunn *et al*. (7)	2019	5	1–77	2/3	3/2	1.1–5.1	AH presents a diagnostic dilemma in imaging and pathology, as it can mimic more malignant lesions, such as angiosarcoma
Yang *et al*. (11)	2022	1	59	Female	Junction of the 2 lobes	10.0	The growth of AH is noninvasive, but the in situ growth can show a large mass effect, which requires aggressive surgical treatment

AH, anastomosing hemangioma.

diagnosis and differentiation of the disease but neglected the growth rate of this tumor (6). The limited available cases showed a slow growth of 5 mm within a 1-year period (7). These limited cases reported interval growth in which the lesion increased in size from 8 to 16 mm over 18 months (8). This case can guide existing understandings of the growth rate of AH in the liver. In our case, AH increased in diameter by 10.7 mm per year, which provides evidence for the study of its biological behavior and offers a reference to guide decisions about the appropriate operation time. The purpose of this case was to demonstrate the deceptive characteristics of MRI images by illustrating the growth potential of a tumor, which necessitates close follow-up in the event that early surgery is deferred.

AH represents a diagnostic dilemma because it has no specific characteristics on CT and MRI images and has overlapping features with angiosarcoma (9). Liver AH is a rare but distinct subtype of hemangioma (10). Only 5 relevant articles have reported cases of liver AH (*Table 1*). A recent study on liver AH showed that its growth is noninvasive *in situ* and requires aggressive resection considering its large mass effect (11). Here, we reported a rare case of a growing AH that mimicked FNH in the liver. Regretfully, we examined gadolinium-enhanced MR imaging, and the MR imaging with the hepatobiliary agent was ignored. FNH was iso- or hyperintense on delayed imaging, which helped to improve the specificity for characterization and diagnosis (12). We observed the characteristic central scar in the lesion, which was not distinct in T1WI or T2WI. The lesion presented as hyperintense in T2WI and hypointense in T1WI and showed no enhancement in arterial (*Figure 2B*) or portal phase but did show delayed enhancement after a 2-minute delay (*Figure 2C*). The typical appearance of the central scar was hypointense on T1WI and hyperintense on T2WI. In one study, hyperintense central scar was demonstrated to be present in 69.1% of cases and in 78% of cases with marked moderate hypointensity on T1WI images (13). Considering the important role of Gd-enhanced T1WI sequences, this case was misdiagnosed as FNH. The MRI demonstrated a lack of aggressive features. When a lesion is diagnosed as benign, the management is primarily surveillance (14). Overwhelming evidence suggests that AH exhibits benign biologic behavior (15). Furthermore, the management and surveillance of AH remain challenging (16). A better understanding of the growth conditions will help guide decisions concerning the optimal treatment of AH (17). Thus, our case report may help settle the controversy concerning whether management should be a surveillance-first or surgery-first approach.

Grossly, the tumor was well-demarcated with a mahogany-brown, solid appearance and a yellow central scar (*Figure 3A*). At higher magnification, the lesions consisted of anastomosing sinusoidal capillary-sized vessels accompanied by scattered hobnail endothelial cells (*Figure 3B*). The corresponding central scar on the MRI images pathologically showed a fibrotic area with few cells and vessels (*Figure 3C*),

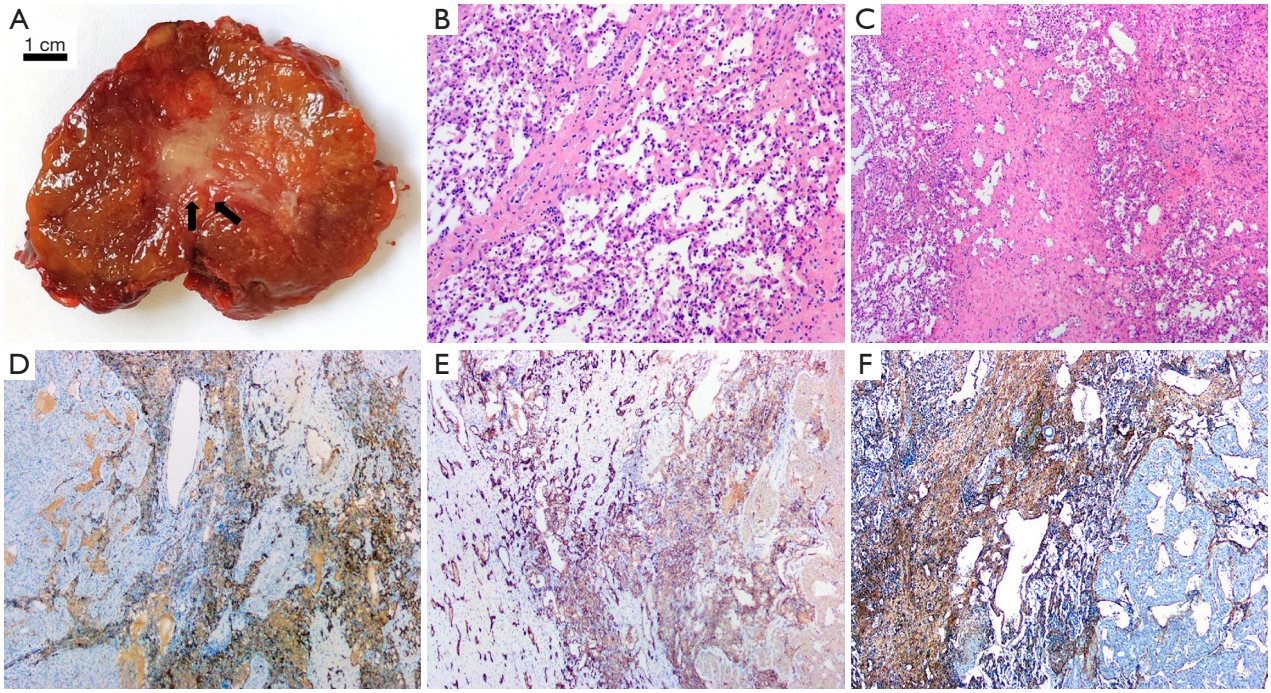

Figure 3 The histopathological images of AH. (A) Grossly, the tumor was a mahogany-brown, solid mass with a yellow central scar (black arrow). (B) Higher magnification showed that lesions consisted of anastomosing sinusoidal capillary-sized vessels with scattered hobnail endothelial cells (HE stain, 300×). (C) The central scar on the MRI images pathologically showed a fibrotic area with few cells (HE stain, 300×). The immunohistochemical stain showed the tumor cells were positive for CD31 (D; immunohistochemistry, 300×) and CD34 (E; immunohistochemistry, 300×). The stromal cells were positive for SMA (F; immunohistochemistry, 300×). AH, anastomosing hemangioma; HE, hematoxylin and eosin; MRI, magnetic resonance imaging; SMA, smooth muscle actin.

which mimicked the fibrous septa, bile ductal proliferation, inflammation, and abnormal blood vessels in FNH (18). This fibrotic area presented as hyperintense on T2WI and hypointense on T1WI images, with the delayed enhancement pattern consistent with the characteristics of fibrous tissue and vessels. The immunohistochemical staining showed the tumor cells were positive for CD31 (*Figure 3D*) and CD34 (*Figure 3E*), and the stromal cells were positive for smooth muscle actin (SMA) (*Figure 3F*).

The differential diagnosis of AH is predominantly with angiosarcoma (19). However, all reported cases of AH lacked mitotic activity, infiltrative growth, or cytologic atypia (20). These factors are easy to distinguish from angiosarcoma (21). We report, for the first time, AH of the liver demonstrating similar radiological features with FNH, including a central scar in the lesion that was pathologically consistent with the fibrous septa in FNH.

In conclusion, liver AH is a rare type of benign hemangioma characterized by anastomosing vessels, hobnailed endothelial cells, benign behavior, and a variety of imaging manifestations. The liver AH demonstrated similar radiological features as those of FNH. This report shows that immunohistochemical stains for CD31, CD34, and SMA can be performed to highlight the vascular nature of the neoplasm. With regard to the probability of the AH growing, for suspicious lesions that do not undergo early surgery, follow-up with appropriate imaging examination should at least be considered.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-22-1082/coif).

The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were conducted in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient to publish this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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