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HEPATOBILIARY

# Anastomosing hemangioma of the liver: a case series

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## Abstract

**Purpose** To report imaging and pathologic features of five pathologically proven anastomosing hemangiomas of the liver (AHL).

**Methods** A retrospective review for AHL was conducted using our institutional database from 6/2004 to 3/2018. Histology proven AHL with radiologic imaging available for review were included. A total of five patients who met our criteria were identified from our institutional database. Computed tomography, ultrasound, and magnetic resonance imaging findings, including location, size, attenuation/signal intensity, enhancement characteristics, and additional imaging data were reviewed. The clinical and pathological data were also reviewed.

**Results** The imaging characteristics of AHL are variable, but features such as peripheral or diffuse hyperintensity on diffusion weighted imaging, arterial hyperenhancement without globular interrupted enhancement, and persistent enhancement without complete filling in the delayed phases were more characteristic of AHL. Imaging also demonstrated a lack of aggressive features.

**Conclusions** AHL present a diagnostic dilemma as they can mimic more malignant lesions, such as angiosarcoma, both on imaging and at pathology. While the imaging characteristics of AHL are variable, there are some features which can help distinguish AHL from other liver lesions. When the diagnosis of anastomosing hemangioma is known, the management of choice is primarily surveillance, as intervention can cause unnecessary morbidity, and no degeneration to malignancy has been identified to date.

**Keywords** Hemangioma · Vascular neoplasm · CT · MRI · Ultrasound

## Introduction

Anastomosing hemangioma is a recently described rare variant of capillary hemangioma with a predilection for the genitourinary system and retroperitoneum [1–3]. These lesions are less likely to be found in other locations such as the liver [1, 4, 5]. In the largest case series of anastomosing hemangiomas to date, liver lesions comprised only 2/32 (6%) of cases [1]. Anastomosing hemangiomas of the liver (AHL) present a diagnostic challenge because they can mimic other more common pathologies on ultrasound (US), computed tomography (CT), and magnetic resonance imaging (MRI), including angiosarcoma and hepatic adenoma in adults [5–8].

AHL is a benign vascular neoplasm, which demonstrates characteristic histomorphologic features. AHL is composed of anastomosing proliferation of small capillary-sized blood vessels. The lesions sometimes show slightly infiltrative growth into the surrounding liver parenchyma; however, an overall circumscribed growth pattern is maintained. The vascular channels are lined by endothelial cells with mild cytologic atypia. One characteristic feature of AHL is endothelial cells with a "hobnail" appearance. There is no mitotic activity or necrosis [5]. Small fibrin thrombi are often seen. Immunohistochemical stains including CD31, CD34, and ERG can be performed to highlight the vascular nature of

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this neoplasm. The most important differential diagnosis of AHL is well-differentiated angiosarcoma. The distinction between the two entities can be challenging due to an anastomosing growth pattern. In contrast to angiosarcoma which shows diffuse infiltration into the background liver parenchyma and marked cytologic atypia, AHL maintains an overall lobular and circumscribed growth pattern. Other key distinguishing features of AHL are mild cytologic atypia and lack of mitotic activity.

Imaging is an important adjunct to diagnosis given the pathology of AHL can also mimic more malignant etiologies such as angiosarcoma [5–8]. While often treated with surgical resection given their nonspecific features and concern for possible malignancy, AHL are benign entities and can be managed with surveillance alone if the diagnosis is known [5]. Imaging features of AHL are not well described in literature. In this case series, we present imaging features of five pathologically proven AHL.

## Methods

A retrospective review for AHL was conducted using our institutional database from 6/2004 to 3/2018. Only biopsy-proven AHL with radiologic imaging were included. A total of five patients who met our criteria were identified from our institutional database. CT, US, and MRI findings, including location, size, attenuation/signal intensity, enhancement characteristics, and other features were reviewed. Patients' age ranged from 22 months to 77 years. There were three males and two females. The clinical, radiological, and pathological data were reviewed.

## Case reports

### Case 1

A 33-year-old otherwise healthy female presented with severe mid-abdominal pain in the epigastrium. As part of her workup, a CT scan was performed that demonstrated a 5.1-cm mass in the right hepatic lobe (Fig. 1). The mass was heterogeneously enhancing with a central non-enhancing component. A subsequent MRI was performed with a hepatobiliary contrast agent. The lesion demonstrated hypointensity on T1-weighted imaging (T1WI), hyperintensity on T2-weighted imaging (T2WI), restriction on diffusion-weight imaging (DWI), heterogeneous hyperenhancement in the arterial phase, persistent hyperenhancement in the portal venous phase, and no uptake of hepatobiliary contrast in the 20-min hepatobiliary phase. After multidisciplinary assessment, it was decided the mass was indeterminate but thought to be most consistent with a hepatic adenoma. The patient underwent surgical resection secondary to large lesion size with right hepatic lobectomy. Pathology demonstrated characteristic histologic features of AHL (Fig. 2).

### Case 2

A 67-year-old woman with low back pain underwent an MRI for further workup. Multiple liver lesions were

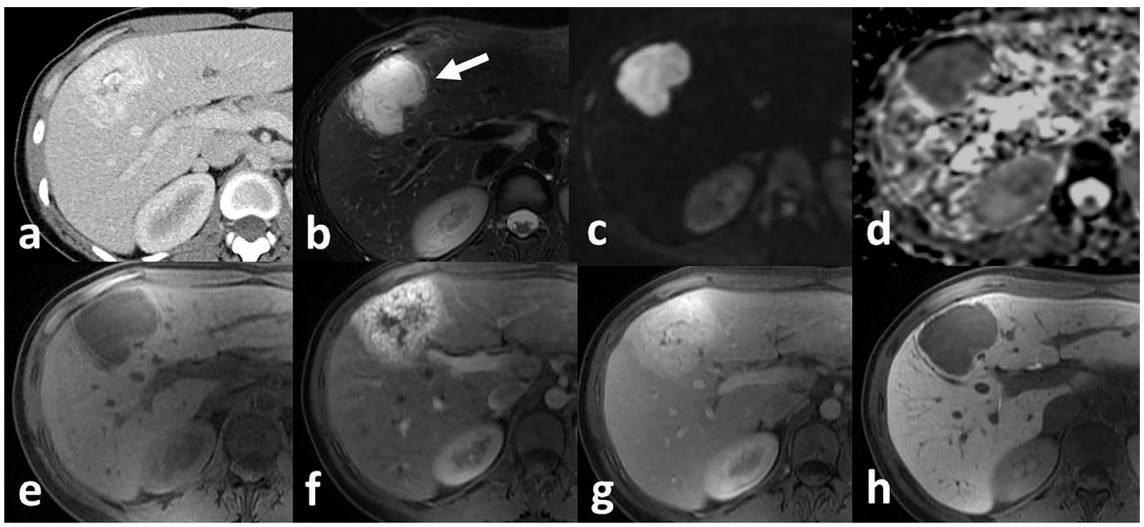


**Fig. 1** Axial contrast-enhanced CT (**a**), T2WI (**b**), DWI (**c**), ADC map (**d**), pre-contrast T1WI (**e**), arterial phase (**f**), portal venous phase (**g**), and hepatobiliary phase (**h**) images demonstrating a large T2 hyperintense lesion in the right lobe of the liver with heterogeneous but predominantly peripheral arterial phase enhancement (arrow) and filling in the portal venous phase. The lesion does not show any uptake with hepatobiliary contrast

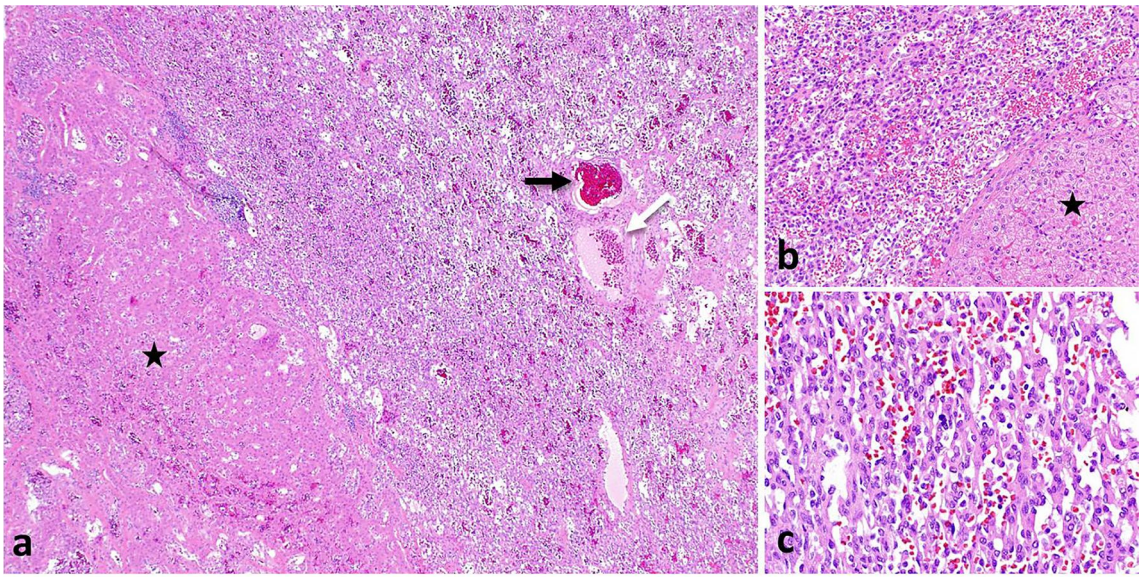


**Fig. 2** Histology of an anastomosing hemangioma (case 1). At low magnification (**a**), the tumor is composed of anastomosing channels and cystic areas containing red blood cells (black arrow) and proteinaceous serum (white arrow). The border of the lesion is well demarcated and clearly circumscribed from the background liver (*) in this resection specimen. (Hematoxylin and eosin; original magnification ×40). Higher magnification (**b**) shows the slit-like spaces are filled with red blood cells. The tumor is sharply demarcated from non-neoplastic liver parenchyma (*) pictured at the bottom right. (Hematoxylin and eosin; original magnification ×200). The endothelial cells lining the vascular channels are slightly atypical, but no mitoses or other features of malignancy are identified (**c**). (Hematoxylin and eosin; original magnification ×400)

incidentally noted, with the largest dominant lesion measuring 1.7 cm. This mass demonstrated isointensity on T1WI, hyperintensity on T2WI, mild restriction on DWI, and solid hyperenhancement in the arterial and portal venous phase (Fig. 3), unlike the peripheral nodular enhancement characteristic of hemangiomas. It did, however, demonstrate persistent enhancement on delayed phase imaging. The patient's alpha-fetoprotein (AFP) level was slightly elevated at 9.2. Given the slightly atypical enhancement and elevated AFP, surveillance was initiated.

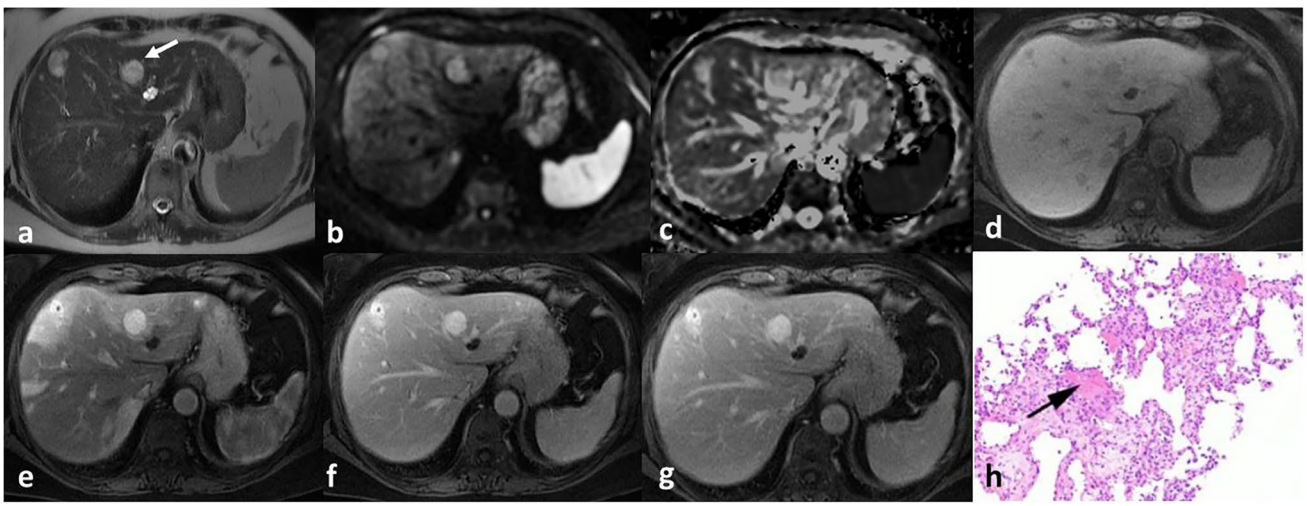


**Fig. 3** Axial T2WI (**a**), DWI (**b**), ADC map (**c**), T1WI (**d**), late arterial phase (**e**), portal venous phase (**f**), delayed phase (**g**) showing multiple T2 bright lesions in the liver that show persistent enhancement in delayed phases. Note the right lobe peripheral lesion shows a persistent filling defect in the center which is thought to represent a thrombus within the lesion. The left lobe lesion (arrow) was biopsied. A needle core biopsy (**h**) showed irregular anastomosing channels lined by endothelial cells. Focal fibrin thrombi (arrow) are present. (Hematoxylin and eosin; original magnification ×200)

Followup MRI showed one of the lesions had increased by 5 mm within a 1-year period and was therefore suspicious. A differential of hepatic adenoma versus metastasis was considered. An image-guided biopsy was performed which revealed AHL. The lesions were followed up with imaging, and at last followup after 3 years, the lesions are stable.

## Case 3

A 77-year-old male with recurrent bloodstream infections had imaging ordered as part of a full workup. A right upper quadrant US demonstrated a 2-cm indeterminate hypoechoic lesion in the right hepatic lobe. CT demonstrated a 2-cm hypervascular ring-enhancing lesion in segment VII of the liver with associated biliary ductal dilatation secondary to compression by the mass. MRI/MRCP showed a 2.1-cm lesion that demonstrated hypointensity on T1WI, hyperintensity on T2WI, peripheral restriction on DWI, and peripheral enhancement with some filling in the later phases. This was favored to represent a healing hepatic abscess given the patient's history of recurrent septicemia (Fig. 4). The patient later presented to the Emergency Department with fever and chills. Infectious disease recommended aspiration of the possible hepatic abscess. On ultrasound, the lesion appeared predominantly solid and an US-guided biopsy was performed with pathology revealing AHL.

## Case 4

A 22-month-old male presented with history of poor weight gain in the setting of chronic loose stools, atopic dermatitis, developmental delay, low immunoglobulin levels, left foot tremors, and a liver lesion incidentally noted on spinal MRI. On ultrasound performed outside, the liver lesion was felt to more likely represent a cyst. Followup ultrasound demonstrated a slight change in size and appearance of the lesion, so an MRI was performed and the lesion was deemed most consistent with a hemangioma or other vascular lesion per outside report. Followup in-facility US demonstrated a macrolobulated, heterogeneous, sonographically indeterminate solid right hepatic mass measuring 1.9 cm. On MRI, the lesion demonstrated hypointensity on T1WI, hyperintensity on T2WI, peripheral restriction on DWI, and peripheral enhancement (Fig. 5). No hepatobiliary contrast agent retention was demonstrated on delayed imaging. In-facility interpretation of the outside MRI felt the lesion was indeterminate. Given the solid nature of the mass on US and MRI imaging, a laparoscopic liver wedge resection was performed and pathology demonstrated AHL.

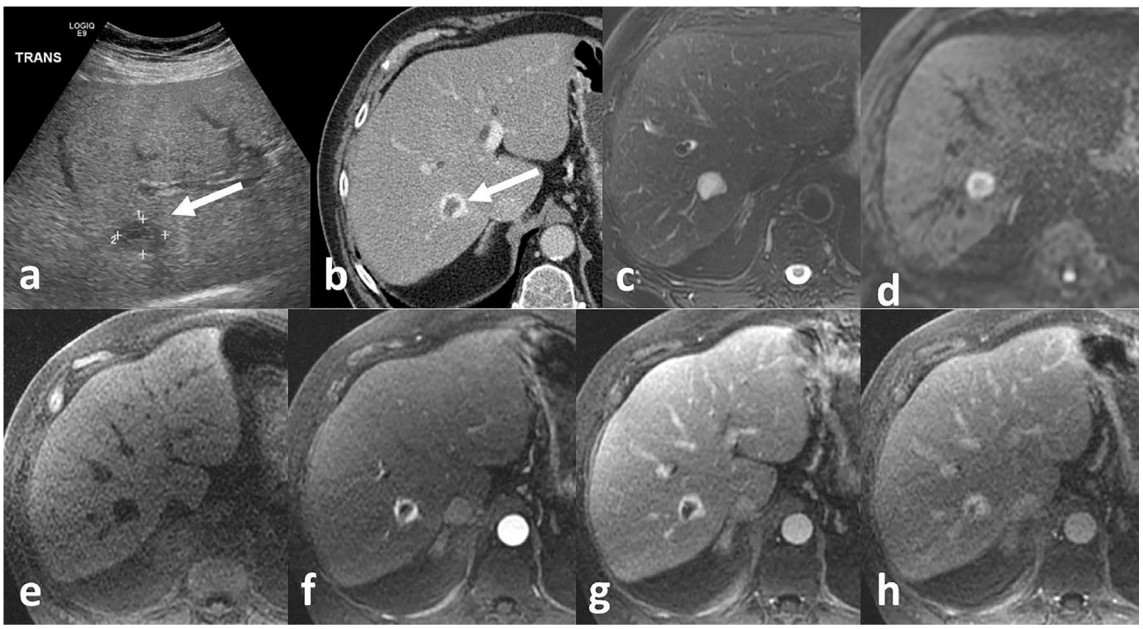


**Fig. 4** Axial ultrasound (**a**) demonstraing a solid appearing hypoechoic lesion in the right lobe of the liver (white arrow). The lesion showing irregular peripheral rim-like enhancement on the portal venous phase CT (**b**). The lesion appearing hyperintense on axial T2WI (**c**) and ring-like hyperintensity on DWI (**d**). The lesion being hypointense on T1WI (**e**) and showing peripheral nearly continuous enhancement in the arterial phase (**f**) that persists and somewhat fills in more during the portal (**g**) and delayed (**h**) phases

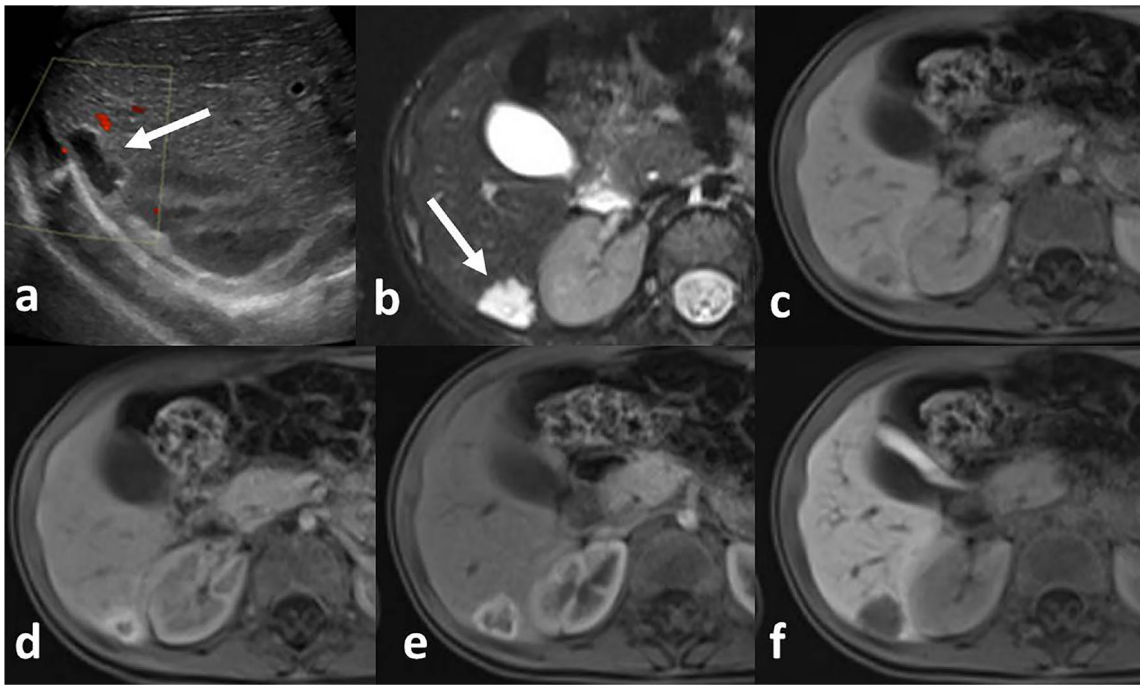


**Fig. 5** Axial ultrasound (**a**) demonstrating a hypoechoic lesion in the right lobe of the liver (white arrow). MRI performed with hepatocellular contrast shows the lesion is hyperintense on axial T2WI (**b**) and hypointense on T1WI (**c**). The lesion shows irregular rim-like enhancement in the arterial phase (**d**) and additional filling in the portal venous phase (**e**). No uptake of hepatobiliary contrast was seen at the 20-min hepatobiliary phase (**f**)

## Case 5

A 48-year-old male with history of mantle cell lymphoma in remission was undergoing surveillance CT for recurrence. A CT abdomen and pelvis with IV contrast incidentally noted a new 1.7-cm peripherally enhancing mass near the dome in the left hepatic lobe. An US of the lesion was performed with findings of a solid mass with a hypoechoic halo concerning for malignancy (Fig. 6). US-guided biopsy demonstrated pathology most consistent with hepatic angiosarcoma (grade II/IV). Given pathology concerning for malignancy, a left lateral hepatic sectorectomy was performed with final pathology demonstrating AHL.

## Discussion

This is the largest case series to date of AHL and the largest series with imaging and pathologic correlates in English literature. All AHL in our series were discovered incidentally. The patient demographics were varied with both males and females represented and ages ranging from 22 months

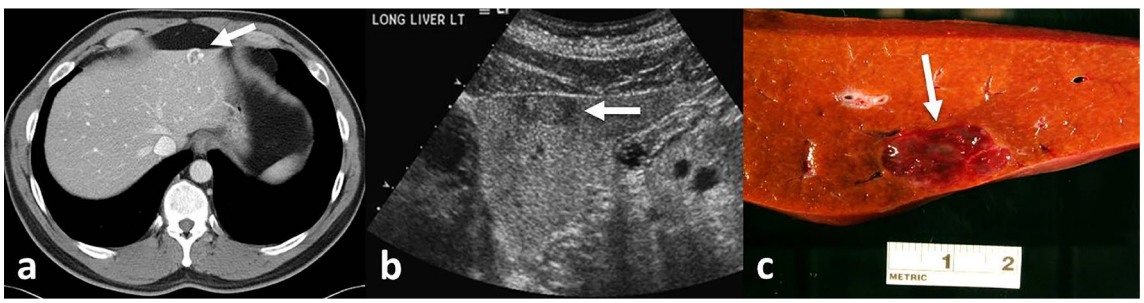


**Fig. 6** Axial contrast-enhanced CT (**a**) demonstrating a rim-enhancing subcapsular lesion in the left lobe of the liver (white arrow). Ultrasound (**b**) demonstrating a hypoechoic rim (white arrow). Resection specimen (**c**) showing the lesion (white arrow)

**Table 1** Imaging features of five pathologically proven AHL

| Case | 1 | 2 | 3 | 4 | 5 |
|---|---|---|---|---|---|
| Age | 67 years | 33 years | 77 years | 22 months | 48 years |
| Sex | Female | Female | Male | Male | Male |
| Reason for imaging | Low back pain | Severe mid-epigastric pain | Recurrent bloodstream infection with unknown source | Left foot tremors and incidental mass in liver on ultrasound | History of mantle cell lymphoma undergoing CT surveillance for recurrence |
| Number of lesions | 1 | 1 | 1 | 1 | 1 |
| Size (AP × trans × SI) | 2.7 × 2.7 × 2.9 cm | 5.1 × 5.0 × 4.9 cm | 2.0 × 1.7 × 2.0 cm | 1.1 × 1.7 × 2.0 cm | 2.7 × 2.4 × 1.7 cm |
| Ultrasound appearance | Solid and hypoechoic with ill-defined margins | – | Solid hypoechoic mass | Macrolobulated, hypoechoic, heterogeneous, solid mass | Solid with hypoechoic halo |
| CT | | | | | |
| Portal phase | – | Heterogeneously enhancing with central curvilinear non-enhancing component | Ring enhancing with central hypoenhancing area | N/A | Solid, peripherally enhancing |
| MRI | | | | | |
| T2W | Hyperintense | Hyperintense | Hyperintense | Hyperintense | – |
| DWI | Mild restriction | Restriction | Peripheral restriction | Peripheral restriction | |
| T1W | Isointense | Hypointense | Hypointense | Hypointense | |
| In/Opp phase | No intralesional fat | No intralesional fat | No intralesional fat | No intralesional fat | |
| Arterial phase | Solid homogeneous | Hypervascular enhancement | Peripheral enhancement | Peripheral enhancement | |
| Portal phase | Persistent enhancement | Heterogeneous solid enhancement | Peripheral enhancement | Peripheral enhancement | |
| Delay phase | Persistent enhancement | – | Persistent peripheral enhancement with some filling in | Lack of retention | |
| Hepatobiliary phase | – | Hypointense | – | Hypointense | – |
| Working diagnosis before pathology | Adenoma or metastasis | Hepatic adenoma | Resolving hepatic abscess or sclerosing hemangioma | Bile duct adenoma or adenoma | Angiosarcoma |

– Denotes no study performed

to 77 years. The lesions had varied imaging appearances (Table 1). On US, the lesions were primarily solid and hypoechoic with variable margins being ill-defined, circumscribed, and macrolobulated. On CT, the lesions had variable enhancement characteristics including heterogeneous, ring, and solid enhancement. On MRI, lesions were iso to hypointense on T1WI with no intralesional fat and hyperintense on T2WI, similar to classic liver hemangiomas. Interestingly, all of them exhibited diffuse or peripheral restricted diffusion. All lesions were hypervascular on arterial phase and showed peripheral thick rim-like enhancement and some filling in the portal venous and delayed phases. However, there was no globular interrupted enhancement in the arterial phase or complete filling in the delayed phases. These features differentiate AHL from the more common cavernous hemangioma of the liver and are consistent with AHL imaging characteristics noted by Peng and colleagues [4]. The lesions did not take up hepatobiliary contrast similar to cavernous hemangiomas. The suspected diagnoses prior to pathologic diagnosis included angiosarcoma, bile duct adenoma, hepatic abscess, and hepatic adenoma.

AHL is a benign vascular neoplasm which displays distinctive morphologic features as compared to other hemangiomas seen in the liver, including cavernous hemangioma and capillary hemangioma. Pathogenesis of this rare entity is unknown. Microscopically, the AHL is composed of anastomosing proliferation of small capillary-sized blood vessels. The lesions usually are well demarcated but sometimes they may show infiltrative growth into the surrounding liver parenchyma. The anastomosing capillary vessels are lined by endothelial cells with mild cytologic atypia which have characteristic "hobnail" appearance. Rare cases of AHL have been described in literature that contained a small component of classic cavernous hemangioma [5]. This finding may

suggest a relationship between AHL and cavernous hemangioma [5]. Anastomosing hemangioma needs to be distinguished from well-differentiated angiosarcoma, which is a highly aggressive neoplasm with a poor outcome. There is some morphologic overlap between the two entities. The main histologic features that distinguish AHL from angiosarcoma are lack of diffuse infiltrative growth, mild cytologic atypia of the lining endothelial cells, and lack of mitotic activity. Attention to these key diagnostic features of AHL can help make a correct pathologic diagnosis.

The imaging characteristics of AHL are variable and can mimic lesions of varying aggressive characteristics, including angiosarcoma, hepatic adenoma, and hemangioendothelioma, and tissue diagnosis is necessary. Imaging is a useful adjunct, however, to demonstrate a lack of aggressive features when absent. Imaging is also useful to potentially differentiate AHL from classic capillary and cavernous hemangiomas, namely with features such as peripheral or diffuse hyperintensity on DWI as compared to no restricted diffusion in classic hemangiomas of the liver, arterial hyperenhancement that does not demonstrate typical globular interrupted enhancement seen in cavernous hemangioma, and persistent enhancement without complete filling in the delayed phases being more characteristic of AHL. The incomplete filling in during delayed phases may be related to frequent presence of thrombi within the lesions. As these lesions can appear solid with moderate T2 hyperintensity, they can be potentially mistaken for other lesions such as hepatic adenoma and hepatocellular carcinoma (HCC). No clinical history of chronic liver disease, absence of capsule, and no washout but persistent enhancement should help to rule out HCC. Epithelioid hemangioendothelioma and angiosarcoma have variable imaging features that can overlap AHL and cannot be reliably excluded on imaging necessitating histological confirmation as was seen in some of our cases. When the diagnosis of anastomosing hemangioma is known, the management of choice is primarily surveillance, as intervention can cause unnecessary morbidity, and no degeneration to malignancy has been identified to date.

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