

Anastomosing hemangioma of liver


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ABSTRACT

Anastomosing hemangiomas are a rare subtype of benign vascular hemangioma which most commonly arise in the genitourinary tract and retroperitoneum. In only a small number of reports has this entity been shown originating within the liver parenchyma. Despite their benign behavior, on contrast-enhanced computer tomography and magnetic resonance imaging studies anastomosing hemangiomas can demonstrate enhancement characteristics similar to primary and metastatic liver lesions. This case report highlights the imaging features of this entity and provides a brief review of the limited literature that exists on this rare hepatic lesion.

CASE REPORT

CASE REPORT

A 56-year-old male with a history of gastrointestinal stromal tumor (GIST) of the terminal ileum and right colon status-post resection 16 years prior presented for additional evaluation following the incidental discovery of a mass within the right hepatic lobe. This mass was discovered on surveillance imaging performed at an outside facility.

Abdominopelvic computed tomography (CT) performed without intravenous contrast (due to history of prior anaphylactic reaction) showed an ill-defined hypoattenuating mass within hepatic segment VII (Fig. 1). Targeted ultrasound imaging yielded a corresponding centrally isoechoic, peripherally hypoechoic lesion with evidence of internal vascularity on Doppler interrogation (Fig. 2). Subsequent magnetic resonance (MR) imaging performed with and without gadolinium demonstrated a slightly lobulated T1 hypointense, T2 hyperintense mass within hepatic segment VII, measuring approximately 3.4 x 3.0 x 2.8 cm, with homogeneous arterial-

phase hyperenhancement which persisted on both portal venous and delayed (8-minute) phase imaging (Fig. 3). No lymphadenopathy or evidence of distant metastases were noted on either examination.

Preoperatively, cavernous hemangioma was suggested as a differential consideration, primarily given T2 characteristics. However, it was noted that the enhancement pattern was atypical for this entity. Given these features, as well as the patient's remote history of GIST, the decision was made to proceed with image-guided biopsy of the liver mass.

Percutaneous biopsy of the lesion showed a circumscribed proliferation of small and thin-walled vascular spaces with hobnailed endothelial cells and without cytologic atypia or mitotic activity (Fig. 4). The strongly and diffusely positive CD34 staining in conjunction with the H&E morphology was overall compatible with anastomosing hemangioma. Importantly, there were no malignant features on pathology nor any findings to suggest recurrent GIST. Given these

findings, the decision was made to monitor the lesion with surveillance imaging rather than undergo surgical resection.

DISCUSSION

Etiology & Demographics:

Anastomosing hemangioma, is a rare subtype of hemangioma only recently described in the literature [1]. Lesions with similar morphology and infiltrative growth arising in the liver have been referred to as hepatic small vessel neoplasms [2-4]. This benign vascular tumor has most commonly been reported within the genitourinary tract and retroperitoneum, although it has been demonstrated to arise within multiple other organs including the adrenal glands, breast, and liver [5-7]. Reports of this entity occurring within the liver are exceedingly uncommon, with only twenty-six documented cases described in the literature to date [2-4, 7-9]. Among the limited cases on record within the liver, no clear trends have emerged regarding any unifying patient demographic features. Patient age at the time of initial diagnosis ranges from 24-83 years (average: 54.9 years). No significant gender predominance appears to exist (15 males; 11 females). No clear lobar predominance exists among hepatic anastomosing hemangiomas. Lesion sizes within the liver vary and have been described up to 16 cm in maximal dimension, which is notably larger than anastomosing hemangiomas documented elsewhere in the body [10].

Clinical & Imaging Findings

A few case series have described the imaging features of anastomosing hemangioma and those which exist have not demonstrated any specific imaging features. Ultrasound characteristics of anastomosing hemangiomas within the kidney have been detailed previously, with lesions noted to demonstrate variable echogenicity [11]. However, no prior reports have described the sonographic features of hepatic anastomosing hemangioma. Notably, the lesion in the present case demonstrates heterogeneous echotexture without definite posterior acoustic enhancement, both features which differ from that of the typical ultrasound appearance of cavernous hemangioma.

In addition to the present report, Peng et al., also described the MR imaging features of anastomosing hemangioma within the liver [8]. This report discusses a hepatic mass found in a 57-year-old woman with no significant past medical history, with the lesion appearing as a rounded well-circumscribed T1 hypointense and T2 hyperintense mass. In this regard, the non-contrast MR findings in the present case appear concordant with this description. However, Peng et al. described their lesion as demonstrating avid peripheral enhancement on arterial phase imaging which persisted on delayed imaging without ever demonstrating central enhancement. In contrast, the lesion in our case showed homogenous arterial-phase enhancement, both peripherally and centrally, which persisted on delayed-phase imaging. The lack of central enhancement seen on the present study is of uncertain etiology, although could potentially relate to the

presence/absence of intralesional thrombus or differences on contrast phase-timing between the two image acquisitions.

While MR findings in the two reported cases are not identical, in both cases the enhancement characteristics were atypical for cavernous hemangioma, which classically demonstrates nodular peripheral enhancement with gradual centripetal filling on delayed-phase imaging. The rapid and persistent homogenous enhancement seen in the present case more closely resembles that of a capillary hemangioma, also known as a fast-flow hemangioma [12]. However, capillary hemangiomas are classically described as smaller lesions (most of which are less than 1 cm in diameter), differing from the comparatively larger sizes noted in the (albeit sparse) collection of reported cases of hepatic anastomosing hemangioma [13].

The diagnosis of anastomosing hemangioma is confirmed by histologic analysis, where it is characteristically composed of an anastomosing proliferation of tightly packed capillary channels with hobnailed endothelial cells [4]. Hepatic small vessel neoplasms show similar morphology with an infiltrative growth pattern [9]. Immunohistochemical stains demonstrate strong and diffuse CD31 and/or CD34 positivity, compatible with a vascular neoplasm. Importantly, there is no mitotic activity, absent or mild cellular atypia, and low Ki-67 index, all features consistent with a benign process [7]. These benign features are crucial to demonstrate, as well-differentiated angiosarcoma, a rare and aggressive malignant tumor, also show hobnailed endothelial cells on histology, and can mimic an anastomosing hemangioma.

Treatment & Prognosis

In all instances on record, hepatic anastomosing hemangiomas were initially discovered as incidental findings on imaging obtained for other purposes [4-8]. At no time in the literature have any patient symptoms been attributed to these lesions, although a single patient demonstrated mild elevation in liver function tests of uncertain etiology [2]. Although anastomosing hemangiomas are considered vascular tumors, importantly, there has never been any documented hemorrhagic complication (either spontaneous or related to percutaneous biopsy) for these lesions seen in the liver or at any other location [7]. Follow-up imaging is available for ten cases of hepatic anastomosing hemangioma, seven of which were treated via partial resection, with no evidence of recurrent lesion or distant metastases noted at follow-up periods up to 96 months post-resection [2, 7]. Given the uniformly benign features of this entity, preoperative diagnosis via percutaneous core biopsy may obviate the need for surgical resection.

Differential Diagnoses:

Cavernous Hemangioma

Cavernous hemangiomas represent the most common histologic subtype of hemangioma and the most common benign hepatic tumor with a reported incidence up to 20% [14]. Histologically, these lesions consist of large vascular

spaces with a central cavernous zone and relatively minimal connective tissue [12]. On US, cavernous hemangiomas typically present as a hyperechogenic, homogenous lesion with posterior acoustic enhancement. On MRI, they present as well-circumscribed round or lobular lesions with homogeneously hyperintense T2 signal with low signal on T1-weighted imaging [15]. Enhancement kinetics are slow, with a classic nodular peripheral enhancement in the arterial phase and gradual centripetal progression to uniform enhancement with persist filling on delayed phase imaging [12].

Capillary Hemangioma

Capillary hemangiomas represent another common histologic subtype of hemangioma and histologically appear as small vascular spaces with extensive connective tissue [12]. These lesions are typically small, with the majority measuring under 1 cm in diameter [13]. On US, they most often appear as hypoechogenic and homogeneous. Non-contrast MRI features of capillary hemangiomas are identical to their cavernous counterparts, with homogeneously T1 hypointense and T2 hyperintense signal characteristics. Enhancement kinetics are where these two entities primarily differ, as capillary hemangiomas classically demonstrate rapid enhancement kinetics characterized by early intense homogenous enhancement in the arterial phase. These lesions are commonly associated with arterioportal shunts, in which transient perilesional enhancement is often appreciated [16].

Primary Hepatic Angiosarcoma

Hepatic angiosarcoma accounts for only 2% of primary hepatic tumors, but is the most common malignant mesenchymal tumor of the liver [17]. Demographically, hepatic angiosarcoma is most commonly seen in elderly patients (50-79 years) and has a known association with exposure to carcinogens, including thorotrast, vinyl chloride, and arsenic [18]. Overall prognosis is quite poor, with early metastases being a common feature and median survival rates ranging from 6 to 12 months [19-20]. Histologically, the anastomosing appearance of hepatic angiosarcoma upon biopsy can mimic that of anastomosing hemangioma. However, the prominent nuclear atypia which characterize angiosarcoma differs from that of the relatively minimal nuclear atypia seen in anastomosing hemangioma [8]. Ultrasound typically shows multiple predominately hypoechogenic lesions of heterogenous echotexture. On MR imaging, hepatic angiosarcoma demonstrates heterogenous T1 signal suggestive of areas of internal hemorrhage. Signal heterogeneity and fluid-fluid levels are often present on T2-weight imaging, additional features which illustrates the hypervascular and often hemorrhagic nature of this entity. Dynamic post-contrast imaging demonstrates heterogeneous enhancement on both arterial and portal venous phases with progressive homogenous enhancement seen on more delayed phase images [17].

Hepatic epithelioid hemangioendothelioma

Hepatic epithelioid hemangioendothelioma is a rare tumor of vascular origin, usually defined as a low-to-intermediate-grade malignancy [21]. On imaging, most lesions are nodular and multifocal with a tendency to coalesce into a conglomerate mass—typically in a peripheral location. Ultrasound can show

discrete nodules or may instead demonstrate diffusely heterogenous echotexture related to extensive involvement. While individual lesions are most frequently hypoechogenic, their echotexture is ultimately quite variable [22]. Non-contrast MRI typically demonstrates the lesions as homogeneously T1 hypointense and heterogeneously T2 hyperintense [23]. Following intravenous contrast administration, these lesions often demonstrate thin peripheral (halo) enhancement, occasionally displaying a targetoid appearance due to the presence of a centrally sclerotic zone with peripheral cellular proliferation [24]. As these are malignant lesions, extrahepatic involvement is a common feature with typical sites of metastatic involvement including peritoneal lymph nodes, omentum, and mesentery [25].

TEACHING POINT

Hepatic anastomosing hemangiomas are a rare and benign subtype of hemangiomas, which demonstrate similar T1/T2 features to their conventional counterpart although possess variable enhancement characteristics that may mimic more aggressive liver lesions on imaging. As imaging cannot definitively distinguish this entity from malignant lesions, histologic confirmation is therefore required. Prior literature as well as the present case suggest this is possible on the basis of percutaneous biopsy, which may obviate the need for surgical resection.

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FIGURES

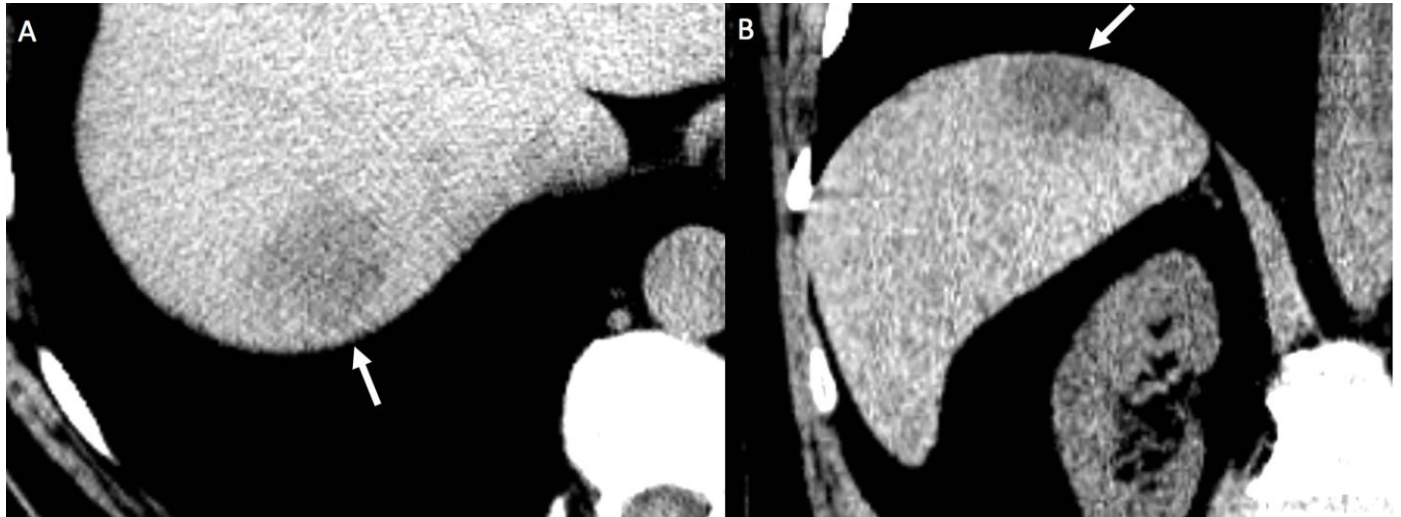


Figure 1: 56 year-old man with hepatic anastomosing hemangioma on noncontrast CT.

Findings: Axial (1A) and Coronal (1B) noncontrast CT images of a homogeneously hypoattenuating lesion (white arrows) with somewhat ill-defined margins within hepatic segment VII. No additional lesions were seen within the liver.

Technique: Toshiba Aquilion PRIME, Scan mode - Helical, Slice thickness - 5mm, mA - 110, kVp - 120, Pitch - 0.81.

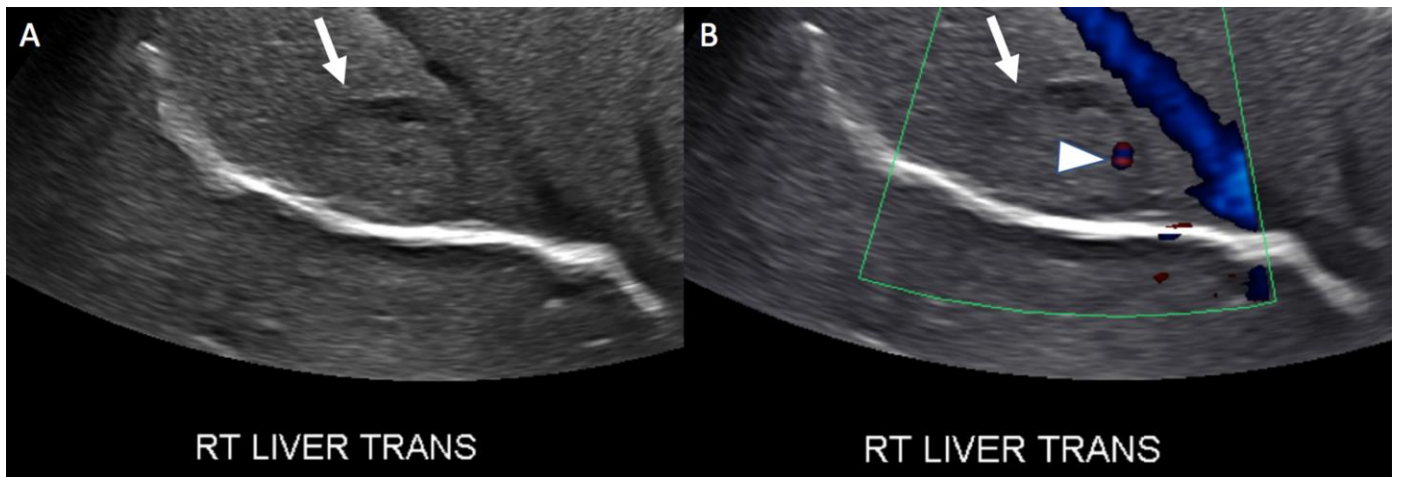


Figure 2: 56 year-old man with hepatic anastomosing hemangioma on ultrasound.

Findings: Transverse (2A) ultrasound images of a centrally isoechoic peripherally hypoechoic lesion (white arrows) within hepatic segment VII. Doppler interrogation (2B) demonstrates a feeding vessel within the periphery of the lesion (arrowhead).

Technique: Toshiba Aplio 500, Transducer - 6Cs1.

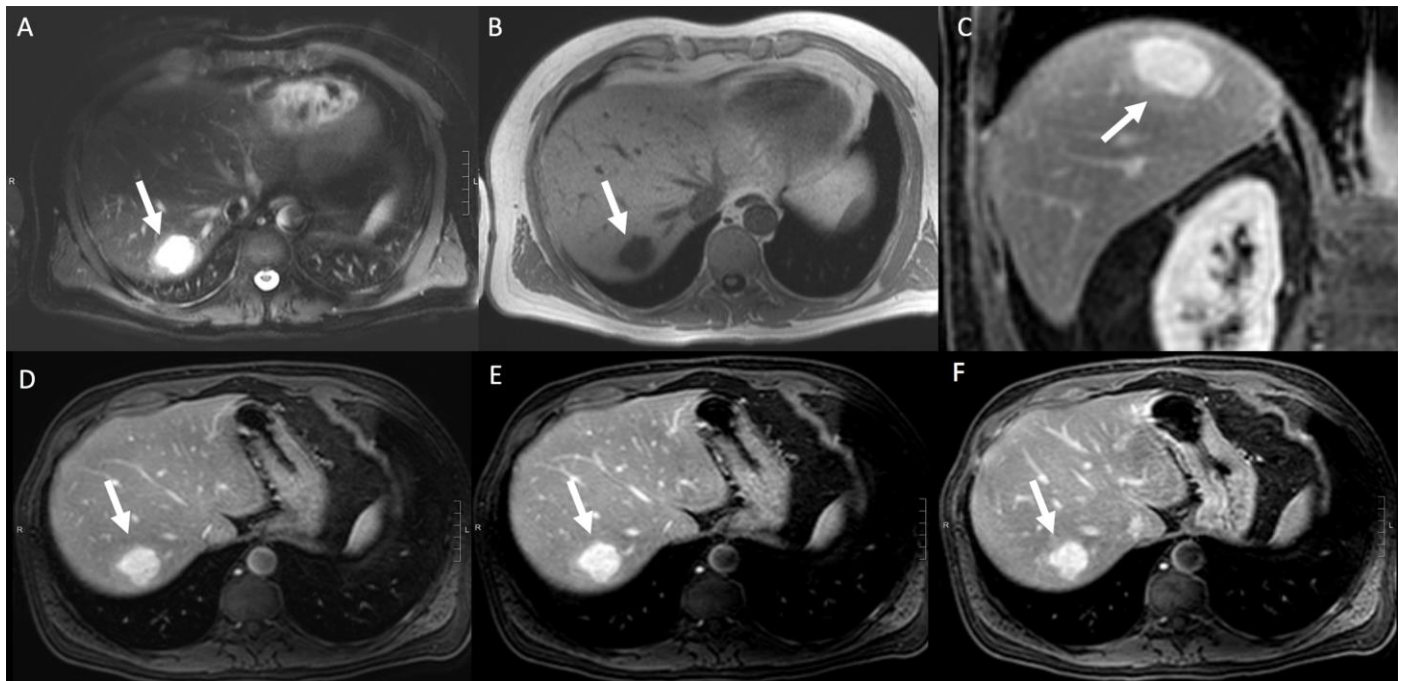


Figure 3: 56 year-old man with hepatic anastomosing hemangioma on contrast-enhanced MRI.

- 3A:**
Findings: Axial T2-weighted image with fat-suppression shows a rounded well-circumscribed homogeneously hyperintense mass in hepatic segment VII (white arrow). No additional liver lesions were identified.
Technique: 3.0 Tesla MRI (Philips Medical Systems Achieva) , TR - 1264, TE - 70, Slice thickness - 7.5 mm.
- 3B:**
Findings: Axial T1-weighted image without fat-suppression shows a rounded well-circumscribed homogeneously hypointense mass in hepatic segment VII (white arrow).
Technique: 3.0 Tesla MRI (Philips Medical Systems Achieva) , TR - 10, TE - 2.3, Slice thickness - 7.5 mm.
- 3C-D:**
Findings: Coronal (3C) and Axial (3D) arterial phase post-contrast T1-weighted image with fat-suppression shows rounded well-circumscribed homogenous intense enhancement within the segment VII mass (white arrows).
Technique: 3.0 Tesla MRI (Philips Medical Systems Achieva) , TR - 2.69, TE - 1.34, Slice thickness - 3.5 mm, Contrast agent - ProHance (Gadoteridol) 0.2 mL/kg.
- 3E-F:**
Findings: Axial T1-weighted post-contrast images in the portal venous phase (3E) and 8-minute delayed phase (3F) demonstrate persistent intense homogenous enhancement without washout (white arrows).
Technique: 3.0 Tesla MRI (Philips Medical Systems Achieva) , TR - 2.69, TE - 1.34, Slice thickness - 3.5 mm, Contrast agent - ProHance (Gadoteridol) 0.2 mL/kg.

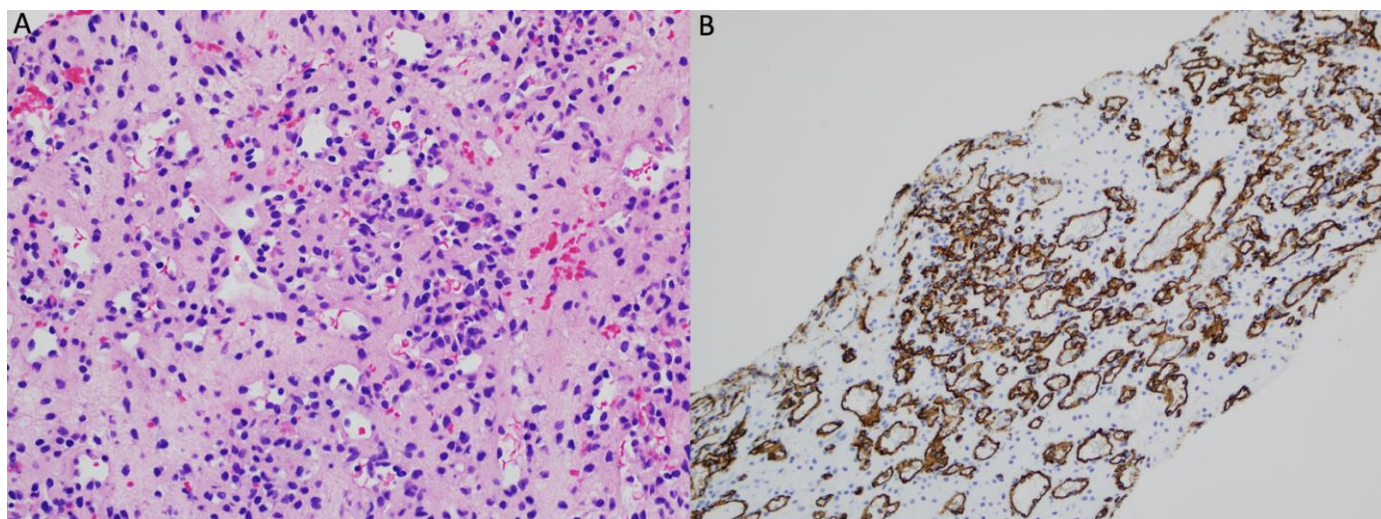


Figure 4: Histopathology findings of hepatic anastomosing hemangioma in a 56 year-old man.

4A:
Findings: H&E sections (magnification: 200X) showed a circumscribed proliferation of small and thin-walled vascular spaces with hobnailed endothelial cells. No cytologic atypia or mitotic activity was seen.

4B:
Findings: Immunohistochemistry (magnification: 200X) for CD34 shows strong, diffuse staining.

Etiology	<ul style="list-style-type: none"> • Unknown
Incidence	<ul style="list-style-type: none"> • Rare (only 26 cases reported in literature as of May 2019)
Gender ratio	<ul style="list-style-type: none"> • Male:Female ratio = 15:11
Age predilection	<ul style="list-style-type: none"> • 24 – 84 years (average age: 54.9)
Risk factors	<ul style="list-style-type: none"> • Unknown
Treatment	<ul style="list-style-type: none"> • Surveillance • Surgical resection
Prognosis	<ul style="list-style-type: none"> • Among cases in which follow-up information is available, there has been no evidence of disease recurrence up to 96 months post-resection
Imaging findings	<ul style="list-style-type: none"> • Well-circumscribed solitary mass • Size range: 0.2 – 16.0 cm (maximal diameter) • T1 hypointense/T2 hyperintense mass • Variable enhancement characteristics

Table 1: Summary table for hepatic anastomosing hemangioma.

	US	CT	MR	Enhancement Pattern
Cavernous Hemangioma	<ul style="list-style-type: none"> • Homogenous, • Hyperechoic, • Posterior enhancement, • Typical diameter < 3 cm 	<ul style="list-style-type: none"> • Homogenous, Hypodense, • Well-circumscribed, • Solitary 	<ul style="list-style-type: none"> • T1: Homogenous, Hypointense • T2: Homogenous, Hyperintense 	<ul style="list-style-type: none"> • Slowly progressive nodular centripetal enhancement
Capillary Hemangioma	<ul style="list-style-type: none"> • Homogenous, • Hypoechoic, • Typical diameter <1 cm 	<ul style="list-style-type: none"> • Homogenous, Hypodense, • Well-circumscribed, • Solitary 	<ul style="list-style-type: none"> • T1: Homogenous, Hypointense • T2: Homogeneous, Hyperintense 	<ul style="list-style-type: none"> • Early diffuse enhancement, • Persists on delays
Hepatic Angiosarcoma	<ul style="list-style-type: none"> • Heterogeneous, • Predominantly hypoechoic 	<ul style="list-style-type: none"> • Heterogeneous, Hypodense, • +/- hemorrhage, • Multifocal 	<ul style="list-style-type: none"> • T1: Variable, depending on hemorrhagic contents • T2: Heterogeneous, Hyperintense, • +/- Fluid-fluid levels 	<ul style="list-style-type: none"> • Heterogeneous enhancement early, • Homogenous enhancement on delays
Hepatic Epithelioid Hemangio-endothelioma	<ul style="list-style-type: none"> • Variable echogenicity, • Diffusely heterogeneous liver echotexture in more extensive forms 	<ul style="list-style-type: none"> • Homogenous, Hypodense, • Multifocal, • Peripheral location 	<ul style="list-style-type: none"> • T1: Homogenous, Hypointense • T2: Heterogeneous, Hyperintense, • +/- Targetoid appearance 	<ul style="list-style-type: none"> • Peripheral enhancement with thin non-enhancing rim
Anastomosing Hemangioma	<ul style="list-style-type: none"> • Peripherally hypoechoic, • Centrally isoechoic, • No posterior enhancement 	<ul style="list-style-type: none"> • Homogenous, Hypodense, • Well-circumscribed, • Solitary 	<ul style="list-style-type: none"> • T1: Homogenous, Hypointense • T2: Homogenous, Hyperintense 	<ul style="list-style-type: none"> • Variable, but in the present case early diffuse enhancement, • Persisting on delays

Table 2: Differential diagnosis table for hepatic anastomosing hemangioma.

ABBREVIATIONS

CD = Cluster of differentiation
 CT = Computed tomography
 GIST = Gastrointestinal Stromal Tumor
 H&E = Hematoxylin and eosin
 MRI = Magnetic resonance imaging
 US = Ultrasound

KEYWORDS

Anastomosing hemangioma; Hepatic small vessel neoplasm; Liver; MRI; CT; US

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