North Clin Istanb 2023;10(4):524–526
doi: 10.14744/nci.2022.21957

Case Report INTERNAL MEDICINE

A rare benign tumor of the liver mimicking angiosarcoma: Anastomosing hemangioma

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ABSTRACT

Anastomosing hemangioma of the liver (AHL) is a very rare condition and limited to a few cases. It is often confused with well-differentiated angiosarcomas and causes overtreatment. In this report, we present a 53-year-old female patient diagnosed with AHL. Since the tumor is rarely seen, it is important to define well the imaging and pathological features for preventing unnecessary surgeries and related morbidities.

Keywords: Anastomosing hemangioma; benign neoplasm; liver.

Cite this article as: *Ismayilov R, Babaoglu B, Keskin O. A rare benign tumor of the liver mimicking angiosarcoma: Anastomosing hemangioma. North Clin Istanb 2023;10(4):524–526.*

Anastomosing hemangioma is a rare benign vascular tumor which first described in 2009 in the genitourinary system [1]. Although it is mostly encountered in the retroperitoneum and kidney, it has also been reported in the adrenal gland, testis, ovary, spermatic cord, mesentery, gastrointestinal tract, and liver [2, 3]. Anastomosing hemangioma of the liver (AHL) is a very rare entity and up to 30 cases have been published in English literature, with no meaningful characteristics such as age, gender, or lobar precedence [4]. The tumor can easily be confused with well-differentiated angiosarcomas in radiological and even histopathological examinations [5]. In this report, we present the imaging and pathological signs of a patient with AHL.

CASE REPORT

A 53-year-old female was directed to our center due to a suspicious liver neoplasm on abdominal ultrasonography of the other center. The patient, who was cholecystectomized for gallstones 5 years ago, had complaints of bloating for 12 years. There was no cigarette smoke or alcohol exposure and a family history of malignancy. No pathological finding was detected in the physical examination. In laboratory analysis, liver function tests were normal, viral hepatitis markers were negative, and the alpha-fetoprotein level was 2.64 ng/mL (normal range, 0–9). Magnetic resonance imaging (MRI) of the upper abdomen showed a 2.5 cm diameter lesion in liver segment 2. The tumor was hy-

The abstract of the article was presented as a poster presentation at the 38th National Gastroenterology Week of the Turkish Gastroenterology Association on 16–21 November 2021.

Received: April 26, 2022 *Revised:* June 17, 2022 *Accepted:* July 04, 2022 *Online:* July 31, 2023

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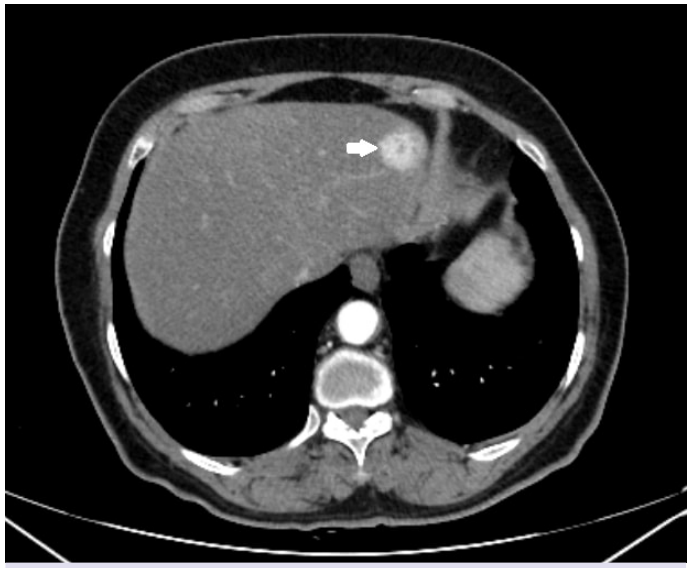

FIGURE 1. Abdominal CT. A 2.5 cm lesion in the left lobe of the liver, with dense contrast-enhancement in both arterial and late phases.

pervascular in the arterial phase and hypodense compared to the liver in the hepatobiliary phase in MRI. Abdominal computed tomography (CT) revealed the lesion enhanced both arterial and late phases (Fig. 1).

Due to the high suspicion of angiosarcoma on imaging, the patient underwent laparoscopic left lobe segment 2 resection. In the histopathological examination, a neoplasm characterized by anastomosis of thin capillary-like vessels and no cytological atypia or mitosis was observed. The neoplastic cells usually revealed the morphology of flattened epithelium and occasional hobnail appearance (Fig. 2). In immunohistochemical studies, neoplastic cells were positive with vascular markers such as CD31, CD34, and ERG. Desmin, Cam 5.2, Heppar, HMB45, and HHV8 were negative (Fig. 3). Ki-67 proliferation index was low. The patient herewith was diagnosed with AHL and the tumor did not recur or metastasized during the 6-month follow-up. Written informed consent for publication of their details was obtained from the patient.

DISCUSSION

AHL is usually incidental and has changeable radiological features that may overlap with angiosarcoma. In the largest series of five patients examining the radiological features of AHL, more characteristic 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 defined, recently [6]. Histopathologically, anastomosing hemangiomas are comprised of tightly packed capillary channels with anastomosing architecture which show hobnail appearance and nonlobular growth pattern imitating well-differentiated angiosarcomas. However, a high grade of cytological atypia and mitotic activity, and the absence of multilayering endothelial cells are the main clues in the differential diagnosis [7, 8].

Since anastomosing hemangiomas can be seen in many intra-abdominal organs, especially in the retroperitoneum and kidneys, imaging should be carefully exam-

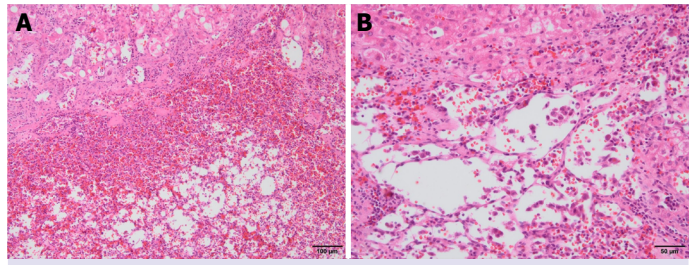

FIGURE 2. Hematoxylin and eosin staining. **(A)** Low magnification reveals multiple capillaries with variable sizes and accompanying congestion. The infiltrative appearance raises suspicion of angiosarcoma. **(B)** At high magnification, anastomosing sinusoidal channels lined by flattened endothelium lacking cytologic atypia and mitosis are observed.

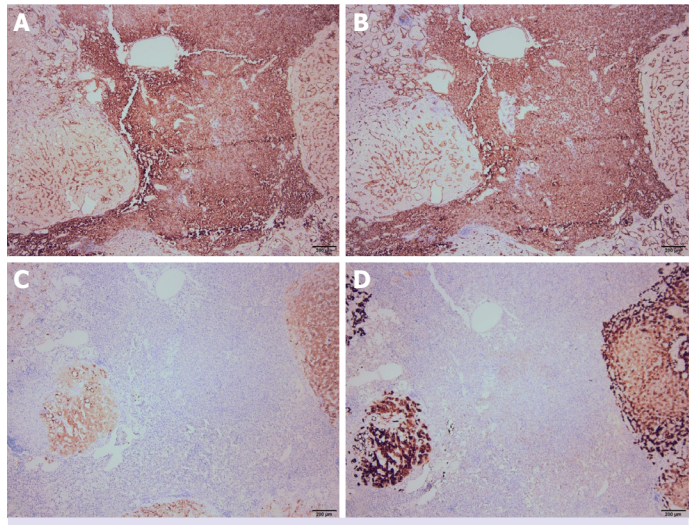

FIGURE 3. Immunohistochemistry. **A** and **B** indicate CD31 and CD34 positive neoplastic cells, respectively. **C** and **D** show Heppar and Cam 5.2 negativity, respectively.

ined in terms of other organ involvement in patients with AHL. In our case, no extrahepatic involvement was detected in abdominal MRI or CT.

No distant metastasis, recurrence, or hemorrhagic complication has been notified in the published cases to date, either after surgical resection or in treatment-free surveillance [4, 9]. Therefore, being aware of this benign tumor and well knowing its radiological and pathological features are of great importance in preventing unnecessary surgeries and related morbidities.

Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

Authorship Contributions: Concept – RI, OK; Design – RI, OK; Supervision – OK; Materials – RI, BB; Data collection and/or processing – RI, BB; Analysis and/or interpretation – RI, BB, OK; Literature review – RI; Writing – RI; Critical review – OK.

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