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Hepatic epithelioid angiomyolipoma is scattered and unsuitable for surgery: a case report

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Abstract

Hepatic epithelioid angiomyolipoma (HEAML) is a rare tumour of mesenchymal tissue with a malignant tendency. Occurring most frequently in women, the relative incidence in men and women, according to incomplete statistics, is approximately 1:5. In rare cases, disease occurrence and development is hidden. Lesions are generally discovered as chance findings by patients; abdominal pain is the first symptom, and imaging has no specificity in diagnosing the disease. Therefore, great difficulties exist in the diagnosis and treatment of HEAML. Here, the case of a 51-year-old female patient with a history of hepatitis B, and abdominal pain over 8 months as the initial symptom, is described. The patient was found to have multiple intrahepatic angiomyolipoma. Due to the small and scattered foci, complete resection was impossible, and because of her history of hepatitis B, conservative treatment was undertaken, with the patient undergoing regular follow-up. When hepatic cell carcinoma could not be excluded, the patient was treated with transcatheter arterial chemoembolization. No tumour neogenesis or metastasis was detected at the 1-year follow-up.

Keywords

Hepatic epithelioid angiomyolipoma, multiple intrahepatic angiomyolipoma, diagnosis, conservative treatment, TACE, regular follow-up

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Introduction

Hepatic epithelioid angiomyolipoma (HEAML) is a rare type of mesenchymal tumour that belongs to the perivascular epithelioid cell tumour (PEComa) group.[1] Epithelioid angiomyolipomas are usually composed of blood vessels, smooth muscles, and adipocytes, and are mostly found in the kidney. More than 50% of angiomyolipomas in the kidney are related to tuberous sclerosis complex; patients with HEAML, however, usually have no liver cirrhosis and HEAML is not accompanied by serological abnormalities.[2] Most patients with HEAML are female, and the lesions are often single, and most are in the right lobe of the liver.[3] The first symptom of HEAML is usually abdominal discomfort, but the majority of patients have no initial symptoms, and lesions are found by chance during regular physical examinations or follow-up examinations for other diseases. At present, the pathogenesis of HEAML remains unclear. Furthermore, imaging examinations lack specificity, the misdiagnosis rate is very high, and the diagnostic accuracy rate is $<32\%$,[4] leading to great difficulties in diagnosis and treatment. The aim of the current study was to present the case of a patient with multiple HEAMLs and discuss the relevant literature published in English. Based on current knowledge and the condition of the patient, the possibility of liver cancer could not be completely ruled out. Therefore, transcatheter arterial chemoembolization (TACE) was continued for treatment and follow-up, to closely observe changes in the patient's condition.

This research was approved by the Ethics Committee of the Shaanxi Provincial People's Hospital, People's Republic of China on 24 February 2022, and written informed consent was obtained from the patient to undergo the procedures described in this report and for its publication. The report was compiled in accordance with CARE guidelines (https://www.equator-network.org/reporting-guidelines/care).[5]

Case report

General information

The present case of HEAML involved a 51-year-old female patient admitted to the Department of Hepatobiliary Surgery, Shaanxi Provincial People's Hospital in September 2020. Her chief symptom was intermittent upper abdominal pain for more than 8 months previously, with no obvious cause. During the previous 8 months, upper abdominal pain had been accompanied by fatigue and poor appetite, but no other clinical symptoms. The patient had not taken her symptoms seriously at first, but after 8 months, the above symptoms occurred repeatedly and with progressive aggravation. She had a history of hepatitis B for 20 years without regular treatment and no history of alcoholism or smoking. Physical examination on admission showed right upper abdomen tenderness, positive percussive pain in the liver area, no liver palm or spider nevus, and no other abnormalities. B-mode ultrasonography suggested an intrahepatic hypoechoic mass with a size of $4.31\,\text{cm} \times 3.97\,\text{cm}$ (Figure 1a). After admission, upper abdominal plain computed tomography (CT) and contrast-enhanced examination showed that the liver was normal in size and shape with smooth edges (Figure 1b–e). A hyperdense shadow was observed during multiple arterial phases (i.e. early and late arterial phases; Figure 1c), but the shadow showed decreased density in late portal phase and delayed phase images (Figure 1d and e), which is similar to that seen in liver cancer. Multiple small, round, low-density shadows with clear boundaries were observed in the right lobe of the liver during the initial CT, with the largest measuring approximately $40\,\text{mm} \times 57\,\text{mm}$. No enhancement was observed on

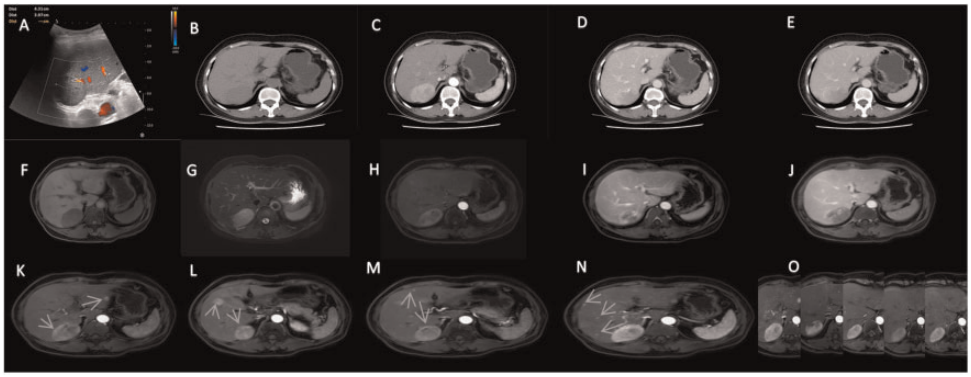

Figure 1. Representative images of various liver tumour manifestations in a 51-year-old female patient: (a) B-ultrasound scan at initial hospital admission showing a low-echoic mass in the liver, approximately 4.13 cm × 3.97 cm in size; (b) plain computed tomography (CT) scan at first hospital admission; (c), (d), and (e) contrast-enhanced CT images showing significantly increased density shadow in the liver parenchyma at early arterial phase, and decreased density shadows at the late portal phase and delayed phase, respectively; (f) and (g) first plain magnetic resonance imaging (MRI) scans showing multiple ovoid and rounded slightly longer T1-weighted and T2-weighted signal shadows in the liver parenchyma; (h), (i), and (j) MRI scans showing obvious uneven enhancement in the arterial phase, but decreased in venous (late portal) and transitional phases; (k), (l), (m), and (n) multiple liver tumours in the mid arterial phase during the first MRI enhanced scan; and (o) MRI images showing tumour (maximum) changes under the capsule of the right posterior lobe of the liver during first hospital admission (September 2020), re-examination and treatment (November 2020), and follow-up (December 2020, March 2021 and September 2021), respectively, showing little change.

contrast-enhanced scan. Multiple abnormal hyperdense shadows in the liver were considered, and HEAML was considered a possibility. Plain magnetic resonance imaging (MRI) scan of the upper abdomen plus diffusion-weighted imaging (DWI) plus enhancement showed multiple ovoid and rounded slightly longer T1- and T2-weighted signal shadows in the liver parenchyma, with clear boundaries and high signal on DWI, and the apparent diffusion coefficient (ADC) value was about $1.38–1.42 \times 10^{-3}\,mm^2/s$ (Figure 1f and g). Enhanced MRI showed obvious uneven enhancement in arterial phase scans, and decreased density regions in the venous (late portal) and transitional phases (Figure 1h, i, and j). No uptake of contrast agent by the lesions was observed during hepatobiliary phase scans. The largest lesion was located under the capsule of the right posterior lobe of the liver, with a size of about 34 mm × 56 mm × 57 mm. A circular long T1-weighted and long T2-weighted signal shadow of approximately 8 mm × 10 mm, with a clear boundary, was observed under the capsule at the top of the liver. The signal was high on DWI, and the ADC value was approximately $3.03 \times 10^{-3}\,mm^2/s$. Multiple small nodules or patchy enhancement were observed in the liver during mid arterial phase MRI scans (Figure 1k, l, m, and n). Due to the presence of multiple abnormal signals in the liver, the following possible diagnoses were considered: (1) HEAML; or (2) hepatic adenoma or other tumours. Laboratory blood test results were as follows: carcinoembryonic antigen (CEA), 0.9 ng/ml; alpha-fetoprotein (AFP), 4.18 ng/ml; cancer antigen (CA) 19-9, 7.6 U/ml; CA 125, 8.24 U/ml; hepatitis B surface antigen (HBsAg) positive; HB e antibody (HBeAb) positive; HBcAb positive;

hepatitis C antibody negative; and normal liver function.

Diagnosis and treatment

The preliminary diagnosis was primary liver cancer stage IIB (China Liver Cancer [CNLC] staging) and stage B (Barcelona Clinic Liver Cancer [BCLC] staging). The analysis suggested that the patient's general condition was stable, the basic condition was normal, and the first consideration was primary liver cancer, although MRI suggested HEAMLs. No pathological biopsy was performed during initial treatment in another hepatobiliary department. According to the patient's medical history and the possibility of multiple lesions suggested by imaging, there was a significant probability of liver cancer. For scattered and multiple hepatocellular carcinoma (HCC), radical resection cannot be achieved by surgery. According to the tumour stage and liver cancer treatment guidelines, transcatheter arterial chemoembolization (TACE) was performed under local anaesthesia to treat multiple lesions, using lipiodol complexed with epirubicin. Intraoperative angiography showed rapid reflux and good development of the portal trunk and left and right branches. The larger lesions were located in the upper posterior segment of the right lobe of the liver, and the left and right hepatic arteries issued branches to participate in blood supply. Multiple nodular neoplastic dense deposits of lipiodol were re-examined after TACE, the tumour supplying artery was occluded, and tumour staining disappeared. Following TACE, further radiography revealed nodular carcinoma of the right posterior upper segment of the liver with multiple intrahepatic metastases.

In November 2020, the patient underwent re-examination by MRI at the hospital, and the size and number of intrahepatic lesions were shown to have undergone little change compared with the previous upper abdominal plain MRI scan plus DWI plus enhancement. After analysis and discussion, it was concluded that, because the number and size of liver lesions had remained relatively unchanged compared with previous scans, TACE treatment was feasible to further control tumour progression. For differential diagnosis and to confirm the pathological diagnosis, B-ultrasound-guided liver puncture biopsy was performed for the largest intrahepatic mass located below the right posterior lobe capsule. Thus, at this visit, liver biopsy was performed and TACE was repeated under local anaesthesia with epirubicin. The remaining lesions were not biopsied because they were too small. Pathological analysis showed that the tumour tissue sample was pale and soft. Microscopically, the tumour was found to comprise epithelioid cells, adipose tissue and blood vessels, with the epithelioid cells arranged in sheets and nests. The cytoplasm was revealed to be eosinophilic. Immunohistochemistry showed that the tumour tissue was AFP negative, CD34 positive, HMB-45 positive, Melan-A positive, and vimentin positive, and the Ki-67 index was approximately 10% (Figure 2). Overall, postoperative pathology showed that the biopsied tumour was a PEComa, which was considered HEAML. Laboratory examination results (CEA, AFP, CA 19-9, and CA 125 levels) were normal, and the patient displayed normal liver function. Based on the patient's medical history, laboratory examination, and imaging results, the patient was diagnosed with multiple intrahepatic HEAML. Considering the large trauma associated with direct surgical resection and the difficulty in retaining an effective liver, surgical treatment was not feasible at this time. Considering the patient's history of hepatitis, and that she was in the basic stage of the 'hepatitis B – cirrhosis – liver cancer' trilogy, new cancer lesions may occur in the liver. After consultation with the patient,

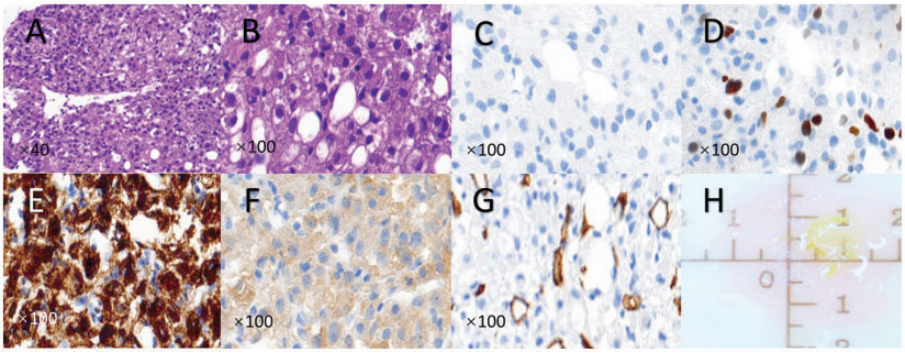

Figure 2. Representative photomicrographs from pathological analysis of liver tumour biopsy tissue from a 51-year-old female patient, showing: (a) and (b) haematoxylin and eosin-stained tumour tissue; (c) negative alpha-fetoprotein staining; (d) Ki-67 index of approximately 10%; positive staining for (e) HMB-45 and (f) Melan-A, suggesting hepatic epithelioid angiomyolipoma; (g) CD 34 positive staining, suggesting a benign tumour; and (h) the pathological specimen at the puncture site; (original magnification stated on each image).

TACE was performed three times in total, and MRI follow-up was conducted approximately every 3 months, in order to monitor disease progression and facilitate further diagnoses in the case of new liver cancer lesions. TACE may also be used to detect lesions missed by CT/MRI. After discharge, treatment with liver-protective drugs (0.5 mg entecavir, orally, once daily and 150 mg diammonium glycyrrhizinate, orally, three times daily) was continued. In addition, hospital follow-up examinations in December 2020, March 2021, and September 2021, by upper abdominal plain MRI scan plus DWI plus enhancement, showed little change in lesions, and no occurrence of new or metastatic lesions, and the largest tumour showed little change during the follow-up (Figure 1o).

Discussion

Perivascular epithelioid cell tumour is an uncommon type of intrahepatic primary tumour, and is more common in adult women than men.[6] Hepatic angiomyolipoma usually exists as a single lesion, most commonly in the right lobe of the liver, followed by the left lobe of the liver, and rarely in the caudal lobe of the liver.[7] Multiple HEAML are rare, and there are few studies on the systemic pathology and prognosis of such lesions. HEAML may be easily misdiagnosed clinically and radiographically as intrahepatic metastasis of HCC. According to the relevant literature, the early drainage vein in HEAML, observed during enhanced MRI, may be the portal or hepatic veins, while the early drainage vein for HCC is only the hepatic vein.[8] In HCC, the blood vessels are mostly connected to the branches of the supplying arteries, but not with veins. AML is mostly connected with the branches of veins, facilitating identification, however, there are few reports about this, and many imaging misdiagnoses are reported in the literature.

Histopathological examinations are effective for differentiating HEAML from HCC, but in a recent multicentre study, histological analysis of liver biopsies revealed misdiagnoses in approximately 15% of cases.[9] By immunohistochemistry, melanocyte markers, such as HBM-45 and Melan-A, are the most specific markers, with positivity for HBM-45 and combination reported in up to 100% of AML cases.[10] In retrospective studies of malignant diseases, Ki-67 has been widely studied as

a potential marker of proliferative prognosis in pathological immunohistochemistry. The Ki-67 index at the critical level of 10–14% positive staining has been associated with a risk of malignancy,[11] while <10% positive staining suggests a malignancy risk of <5%.[12] AFP has been widely used as a tool for HCC screening, diagnosis, monitoring, recurrence monitoring, and prognosis prediction.[13] Although HEAML is benign, it may also be malignant.[14] Therefore, HBM-45, Melan-A, AFP, and Ki-67 indices in immunohistochemistry can be used to differentiate HEAML from HCC, and to differentiate benign from malignant HEAML.

For a single hepatic angiomyolipoma >5 cm in diameter, which cannot be confirmed by imaging and needle biopsy, surgical resection should be performed if there are no obvious surgical contraindications.[13] However, for the treatment of rare tumours with relatively small diameters, and scattered multiple tumours, as in the current case, and as reported previously for a patient with multiple non-hepatitis B-related lesions, treatment should be followed-up by needle biopsy.[1,15] In addition, three patients with multiple non-hepatitis B lesions, reported by Tan et al.,[16] Chai et al.,[17] and Durczyński et al.,[18] underwent segment resection (Table 1). For one patient, the follow-up was unclear. Very few cases of multiple HEAML were found in the previously published literature, and none of the four patients described in Table 1 had hepatitis B. Therefore, the published literature was reviewed for reports of relevant HEAML cases over the past decade (2012–2022). Among 105 patients reported in the literature, 12 patients had hepatitis B virus (HBV), and two had hepatitis C virus infection (Table 2).[1,6,12,16–46] Few patients were found to have HEAML complicated with HBV. However, hepatitis B-positive patients with HCC are quite

Table 1. Summary of cases with multiple liver tumours reported in the literature over the last decade.

Publication		Case									
Reference	Author, year	Age, years	Sex	Tumour size, cm	Location	Starting symptoms	Imaging diagnosis	Region/ Country	Viral hepatitis	Treatment	Recurrence/ metastasis (n)
1	Agaimy, 2012	21	M	Multiple (×8)	Multiple	No symptoms	HCC pattern	European	No	Core needle biopsy	Recurrence, slight progression on imaging
16	Tan, 2012	41	F	Multiple	Left lobe	No symptoms	Unclear lesion	Asian	No	Segment resection	12 mo, ANER or new tumours
18	Durczyński, 2012	18	F	15.0/3.0	Left lobe	Abdominal pain	HCC pattern	European	No	Segment resection	65 mo, ANER or new tumours
17	Chai, 2020	51	F	4.6/1.0	Right lobe	Thoracic spine pain	HCC pattern	Australia	No	Segment resection	Not available

M, male; F, female; HCC, hepatocellular carcinoma; ANER, alive with no evidence of recurrence; mo, months.

Table 2. Summary of cases of hepatic angiomyolipoma with or without concomitant hepatitis B infection reported in the published literature over the past decade (2012–2022).

Type	Number
Sex	
Female	78
Male	26
Not reported	1
Number of tumours	
Single	99
Multiple	5
Malignant tumour	1
HBV	
HBV	10
HBV, HCV	2
No	94
Total	105

HBV, hepatitis B virus; HCV, hepatitis C virus.

common, so the diagnosis and treatment of patients requires care to avoid misdiagnosis.

In the present case, the patient had normal liver function, was negative for tumour markers, and positive for HBsAg, HBeAb, and HBcAb. The patient was also a carrier of the hepatitis B virus. Surgical treatment has disadvantages, such as a large resection range, incomplete resection, and multiple operations, thus, better treatment methods are needed for patients with hepatitis B compared with those without hepatitis B. The current patient was diagnosed with HEAML by biopsy using haematoxylin and eosin-stained sections and immunohistochemistry. Although she was a carrier of hepatitis B, her liver function was normal, without metastasis or new occurrence. The patient had good medical compliance. No abnormalities were found in liver function or tumour changes on intensive MRI. Through MRI monitoring, if malignant lesions are found, they may be diagnosed and treated early.

In patients with hepatitis B, the identification of HEAML and HCC is relatively difficult. According to the Chinese Society for Clinical Oncology 2018 guideline for diagnosis and treatment of primary liver cancer, affected by many factors, such as the lesion size and location, there is a high false negative rate in pathological diagnosis of liver lesions, particularly for lesions with diameters less than 2 cm. Therefore, the negative results of liver biopsy cannot completely rule out the possibility of liver cancer. In China, where the incidence of liver cancer is high, the risk of conservative treatment is high, and hepatitis B carriers do not satisfy the indication for surgery. TACE is one of the most commonly used methods in non-surgical treatment of liver cancer. We believe that TACE plus follow-up may be used to treat patients with multiple HEAML and a history of hepatitis B. Some tumours are too small for pathological biopsy, and TACE may prevent HCC. Because such cases are scarcely reported, with few related guidelines and treatments, the present case report may provide relevant diagnosis and treatment ideas. Future updated HEAML guidelines with improved relevant treatment options for multiple lesions would allow treatment to be adjusted to a greater extent. In the meantime, we can do our best to alleviate patients' pain until the patient can be clinically cured.

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Author contributions

Si-yuan Pan collected patient data and drafted the initial manuscript. Shu-chuan Sun and Jiang-xin Liu conceived and designed the present study. Xiang-reng Cai revised the manuscript along with Si-yuan Pan, Xiang-reng Cai, and Yu-xin Deng, and these individuals were

responsible for the patient's treatment. All authors read and approved the manuscript.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

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