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Hepatic Sclerosing Hemangioma Mimicking Malignancy: A Case and Literature Review

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Abstract

Background: Sclerosing hemangiomas of the liver are rare, benign tumors with degenerative changes. These degenerative changes, however, often obscure the true, benign nature of the tumor and give them features indistinguishable from other malignant processes, thus making the diagnosis difficult.

Case presentation: A 70-year-old male without any previously diagnosed liver disease or malignant process presented with incidental right hepatic mass in ultrasonography and weight loss. Physical exam was unremarkable. The labs were significant for mild pancytopenia, elevated total bilirubin and slightly decreased transferrin. Follow-up triple phase-contrast CT scan of the abdomen revealed a lobulated, poorly demarcated lesion measuring 4.8 x 4.5 cm, located in segment V of the liver with encasement of the left portal branch. The overall picture was indeterminate but highly suspicious for malignancy. A decision was made to perform a CT-guided biopsy which revealed sclerosing hemangioma of the liver.

Conclusion: It is challenging to differentiate sclerosing hepatic hemangioma from atypical hepatocellular carcinoma, intrahepatic cholangiocarcinoma, and metastatic tumors utilizing only imaging modalities. The diagnostic workup should include biopsy of the atypical liver lesion which unveils the final diagnosis and avoid subjecting the patient to an extensive, and invasive surgical resection.

Keywords

hemangioma; sclerosing; liver; imaging features

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1. Introduction

Hemangiomas are the most common benign hepatic lesion with a prevalence ranging from 1% to 20%, as have been documented in autopsies [1,2]. They are typically discovered incidentally on abdominal imaging in patients between the ages of 30 to 50 years old with a female predominance [3,4].

Sclerosing hemangiomas are a rare and benign subtype of this group of tumors. They are caused by degenerative changes such as thrombus formation, fibrosis, and scarring in hepatic cavernous hemangiomas, although the underlying precipitating factor for the degeneration of hepatic cavernous hemangiomas has been unclear [2,5,6].

Hemorrhages, hemosiderin deposition, and mast cells can be seen in sclerosing hemangiomas. While fibrosis, elastic fibers, and dystrophic calcifications with a decreased number of mast cells are observed in completely sclerosed hemangiomas [7].

Whereas non-sclerosing hemangiomas may be easily identified, the sclerosis and other changes seen in sclerosing hemangiomas of the liver obscure the true nature of the lesion, thus making them difficult to diagnose [2]. Furthermore, their atypical radiologic findings often make them indistinguishable from other lesions such as atypical hepatocellular carcinoma, intrahepatic cholangiocarcinoma, and metastatic tumors, therefore often create a diagnostic dilemma.

2. Case Presentation

A 70-year old Caucasian male presented to the clinic for workup of an incidental right lobe hepatic mass (5.2 x 3.9 cm) found on ultrasonography as part of a pancytopenia work up. The patient's past medical history included hypertension, hyperlipidemia and alcohol use disorder. He had no known history of hepatocellular disease or malignancy. The family history was also negative for liver disease or malignancies. He admitted to drinking two wine spritzer three times a month as well as 1-2 alcoholic drinks per day for the past 50 years; he denied any past or current drug use. A review of systems revealed an unintentional weight loss of 14 pounds over the previous 3 months. Vital signs were within normal limits. The physical exam was unremarkable with no stigmata of hepatic disease noted. Complete blood count revealed mild pancytopenia with WBC 4.3 K/Cmm (normal range: 4.5-10.0 K/cmm), hemoglobin 12.7 gm/dl (normal range: 14-17.5 gm/dl) with normal MCV and RDW and platelets 128 K/cmm (normal range: 150-450 K/cmm).

Liver function tests were normal except for a mildly elevated total bilirubin 1.7 mg/dL (normal range 0.1-1.1mg/dL). Tumor markers including alpha-fetoprotein, protein induced by vitamin K absence or antagonist-II, carcinoembryonic antigen, and carbohydrate antigen 19-9 were all unremarkable. Human Immunodeficiency virus, Hepatitis A, B and C serologies were negative. Alpha-1 antitrypsin and ceruloplasmin levels were within normal limits. Serum iron saturation was low (19%) however serum ferritin was within normal limits. Antinuclear antibody and anti-smooth muscle antibodies were unrevealing. Eight months prior, a colonoscopy performed at another institution 8 months prior to presentation, had been remarkable only for one polyp with biopsy showing tubular adenoma.

A triple phase-contrast computed tomography scan of the abdomen revealed a lobulated, poorly demarcated lesion measuring 4.8 x 4.5 cm, located in segment 5 of the liver with encasement of the left portal branch, without clear washout in portal venous phase [Figure A]. The overall radiologic impression was indeterminate with a high suspicion for malignancy. To reach a definitive diagnosis, a CT-guided biopsy of the liver lesion was performed, from which histopathological examination showed foci of prominent fibrosis and hyalinization with narrowed and obliterated vascular spaces consistent with a sclerosing hemangioma [Figure B]. No further evidence of a malignant neoplastic process was noted. Thus, the patient was discharged with the diagnosis of sclerosing hemangioma of the liver and it was determined that no further surveillance was indicated.

3. Discussion

Hepatic sclerosing hemangiomas were first documented by Shepherd and Lee in 1983 [8]. Since then, only a handful of case reports have been published on the topic. The documented prevalence of hepatic sclerosing hemangiomas ranges from 1% to 20% [1,2]. Whereas typical liver hemangiomas most commonly occur in patients between the ages of 30 to 50 years old with a female predominance, hepatic sclerosing hemangiomas most commonly occur between the ages of sixty to seventy years of age, with approximately two thirds of cases occurring in men [3,4]. They are caused by degenerative changes such as thrombus formation, fibrosis, and scarring in hepatic cavernous hemangiomas, although the underlying precipitating factor for the degeneration of hepatic cavernous hemangiomas has been unclear [2,5,6]. Clinical presentation varies from asymptomatic and discovered incidentally on abdominal imaging to symptomatic [3,7]. The most common presenting symptoms in symptomatic cases included abdominal mass and pain [7].

Hepatic sclerosing hemangiomas, although benign, have many features resembling that of other malignancies, which makes the diagnosis and workup challenging. Diagnosis and workup include abdominal imaging to detect the lesion followed by biopsy and/or surgical resection for definitive diagnosis. Surveillance imaging may also aid in the diagnostic workup. Because of its rarity and atypical radiologic findings, sclerosing hemangiomas can be indistinguishable from other liver lesions and as such atypical hepatocellular carcinoma, intrahepatic cholangiocarcinoma and metastatic lesions must also be included in the differential diagnosis. The malignant possibilities in the differential diagnosis make correct diagnosis of the liver lesion so important.

Similar to typical hemangiomas the radiologic features of sclerosing hemangiomas include the presence of a transient hepatic attenuation difference, rim enhancement, and nodular regions of intense enhancement. In contrast however, they could also show geographic pattern, capsular retraction, and loss of previously seen regions of enhancement [9]. The radiological features of sclerosing hemangiomas revealed by dynamic CT and MRI are similar to those of hepatic malignancies making the diagnosis highly challenging based on imaging alone [10]. It is hypothesized, however, that the use of [18F]-fluorodeoxyglucose positron emission tomography (FDG-PET) could be helpful in the preoperative diagnosis to distinguish benign sclerosed hemangioma from a malignant tumor such as intrahepatic cholangiocarcinoma or metastatic liver cancers [11].

Furthermore, being as hepatic sclerosing hemangiomas are benign, once diagnosed, they warrant no further treatment and offer a very good prognosis. The current case demonstrates that despite the high suspicion for malignancy, the tumor was a benign, incidental finding warranting no further workup.

Workup of liver lesions of unknown etiology, such as a hepatic sclerosing hemangioma include biopsy, surgical resection, and surveillance imaging. There is limited data on the diagnostic approach to hepatic sclerosing hemangiomas with an equally limited number of case studies published on the topic. There are multiple cases reported that patients with suspicion for malignancy underwent laparoscopic or open surgery with post-operative pathology revealing benign sclerosing hemangioma [2,12,13]. A common denominator in all these previously reported cases is that the patient was subjected to what ended up being an unnecessary, invasive surgery to resect a lesion that turned out to be benign.

To our knowledge, there is only one previously published case report on the subject for which the patient was not subjected to surgical resection. Behbahani et al. were presented with a case for which the presentation was suspicious for gastrointestinal malignancy and imaging worrisome for hepatic metastasis and triple phase CT showing peripheral heterogeneous enhancement. They performed image-guided biopsy which revealed benign sclerosing hemangioma [10]. This approach is similar to our own in that by opting for an image-guided biopsy as opposed to escalating directly to surgery, a diagnosis was made in the most minimally invasive way possible and spared the patient from an extensive surgery.

Sclerosing hemangiomas have an excellent prognosis. They remain stable for long periods and can be followed without intervention. However, it is extremely difficult to precisely diagnose them solely from imaging modalities. If the possibility of malignancy cannot be ruled out or in cases of ambiguous diagnosis, a targeted biopsy can be used to reach a conclusive diagnosis in the most minimally invasive manner possible.

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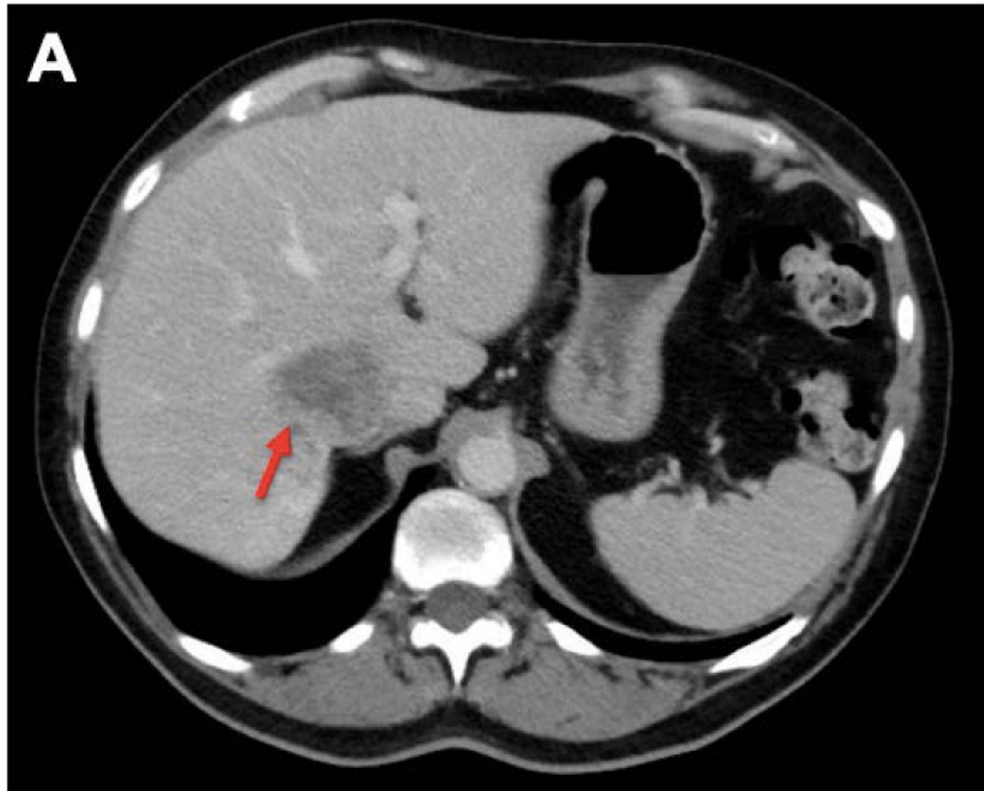


Figure A. Portal venous phase of contrast CT scan showing a lobulated, poorly demarcated liver lesion (red arrow) without clear washout and partial nodular ring enhancement

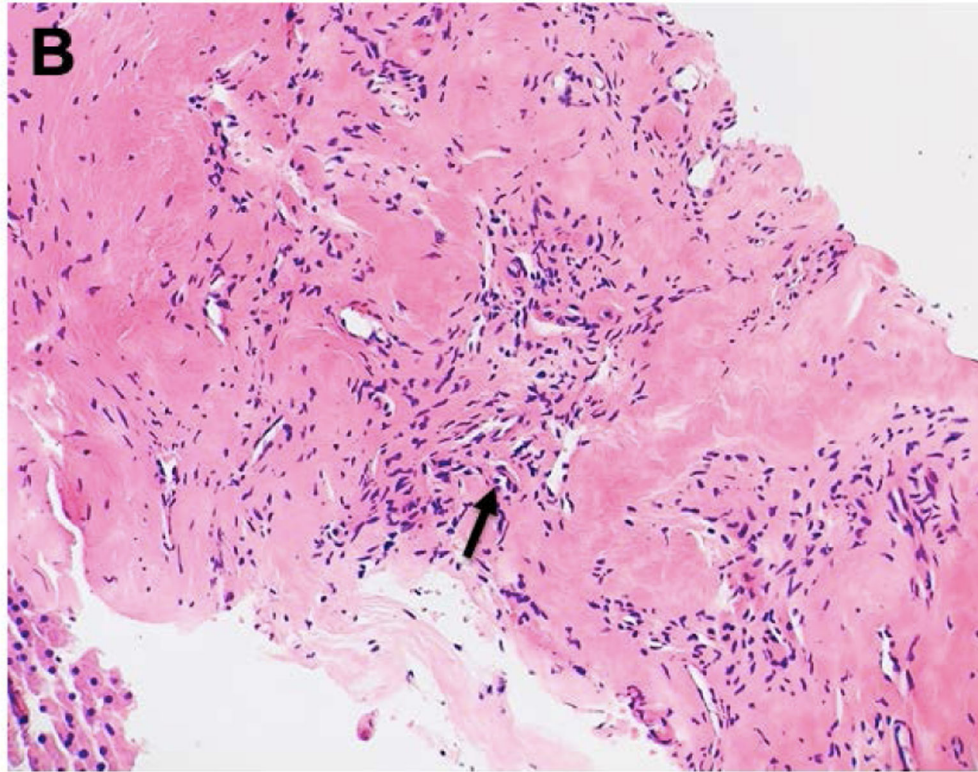


Figure B. Histologic sections of the liver biopsy showing prominent fibrosis and hyalinization with narrowed and obliterated vascular spaces (black arrow) consistent with a sclerosing hemangioma

Table 1.

Laboratory Data

Serum	Patient	Ref. Range
WBC (K/cmm)	4.3	4.5-10.0
RBC (M/cmm)	4.2	4.5-5.9
Hemoglobin (gm/dL)	12.7	14-17.5
Hematocrit (%)	38.2	41.5-50.4
MCV (pg)	91	80-96
RDW (%)	13.6	11-14.5
Platelets (K/cmm)	128	150-450
Sodium (mEq./L.)	144	135-147
Potassium (mEq./L.)	4.5	3.3-5.1
Chloride (mEq./L.)	107	98-112
CO2 (mEq./L.)	26	22-31
Glucose (mg/dL)	93	70-115
BUN (mg/dL)	17	6.5-23
Creatinine (mg/dL)	1	0.6-1.5
Total Protein (g/dL)	7.3	6.4-8.6
Albumin (g/dL)	4.5	3.9-5.2
AST (Unit/L)	15	5-40
ALT (Unit/L)	11	7-45
Alk Phos (Unit/L)	70	30-136
Total Bilirubin (mg/dl)	1.7	0.1-1.1
Ferritin (ng/ml)	257	22-322
Iron (mcg/dl)	75	40-155
TIBC (mcg/dl)	338.2	228-428
Iron Saturation (%)	22.1	20-50
Vitamin B12 (pg/mL)	561	211-911
Folate (ng/mL)	21	5.39-24.00
TSH (mcUI/mL)	1.21	0.45-4.95