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Benign metastasizing pleomorphic adenoma in liver mimicking synchronic metastatic disease from colorectal cancer: a case report with emphasis on imaging findings

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Abstract

Pleomorphic adenoma of the parotid gland with metastases to the liver is a rare etiology of focal liver lesions, and there are no described pathognomonic imaging features. We report a patient who presented with a newly diagnosed rectal cancer and multiple cystic liver lesions suspicious of mucinous synchronous liver metastases. Following chemotherapy no reduction in the number or size of the liver lesions was observed. The patient was re-evaluated and a biopsy of a lesion was performed. The specimen showed a metastasis from a pleomorphic adenoma of the parotid gland for which the patient had been treated 20 years earlier. The case illustrates how a thorough medical history can be crucial when a standard diagnostic imaging workup for colorectal cancer metastases is uncertain, and how a biopsy, though regarded as contraindicated due to the risk of tumor cell dissemination, can be required to secure a correct diagnosis.

Keywords

Pleomorphic adenoma, benign, cystic liver metastasis, ultrasound, computed tomography (CT), magnetic resonance imaging (MRI)

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Introduction

Benign metastasizing pleomorphic adenoma (BMPA) is a rare condition that occurs in patients with a prior history of pleomorphic adenoma (PA) (1,2). PA is the most common neoplasm of the salivary glands and is generally considered a benign tumor. By definition, BMPA is characterized by the presence of one or more foci of histologically benign pleomorphic adenoma outside the salivary glands. Most metastases from PA occur in patients that have been surgically treated one or more times, and a hypothesis is that the tumor spreads by vascular implantation of tumor cells during the surgical procedure, followed by hematogenic dissemination most commonly to bone, but also to the head, neck, and lung (1,2). There is often a long time interval, reportedly up to 51 years (1), between the diagnosis of a primary pleomorphic adenoma and the detection of metastases, and a prior history may be crucial in suggesting the correct diagnosis.

Hepatic metastases are extremely rare and we have found only one case report on imaging of BMPA to the liver (3), and to our knowledge, the imaging characteristics of multiple BMPA of the liver have previously not been described.

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Case report

A 65-year-old woman with no previous history of malignant disease was referred to our university hospital with a newly diagnosed invasive adenocarcinoma of the rectosigmoidum, stage III. A contrast-enhanced computed tomography (CT) scan of the abdomen revealed one sub-capsular lens-shaped hypodense lesion with a maximum diameter of 6.0 cm in segment VII, and five smaller lesions of 0.5–2.0 cm scattered in the parenchyma (Fig. 1). The smaller lesions were oval and/or lobulated in shape, some with irregular outlines. The lens-shaped lesion had pre-contrast attenuation in the range of 30–36 HU, while the 1.0–2.0 cm lesions were 23–44 HU (Fig. 1a), indicating a content of protein-rich fluid of all six lesions. Following injection of intravenous contrast none of the lesions showed enhancement in the arterial (Fig. 1b), the portal venous (Fig. 1c), or the late phase (Fig. 1d). To further evaluate whether the lesions could represent hemorrhagic/inflammatory cysts or mucinous/necrotic metastases from the rectosigmoid tumor, both ultrasound (US) and magnetic resonance imaging (MRI) were performed.

An US of the liver with and without intravenous contrast (SonoVue®) showed hypoechogenic, lobulated, and/or oval lesions with slightly irregular borders and with posterior echo enhancement indicating fluid content (Fig. 2a). Except for the smallest lesions, central septations were displayed. No convincing contrast enhancement was observed (Fig. 2b). The lesions were interpreted as "complicated cysts" or metastases with necrosis or a mucinous content, and to be in line with the CT findings.

To evaluate the presence of additional lesions an MRI exam using the liver specific MRI contrast agent super-paramagnetic iron oxide (SPIO) was performed. SPIO accumulates in the reticuloendothelial system (RES) of the liver, while the cells of most malignant liver lesions do not contain RES. The exam detected another six sub-centimeter lesions; all 11 lesions showed high signal relative to liver parenchyma on T2-weighted (T2W) sequences (Fig. 3a), and low signal on T1-weighted (T1W) sequences, indicating high water content. In addition the MRI confirmed the previous imaging findings of a cystic, lobulated appearance with central septations. Accordingly the suspicion of cystic metastases with necrosis or mucin content were maintained (Fig. 3b).

The patient was discussed at the liver and rectum cancer multidisciplinary teams, which both concluded that the lesions most likely represented mucinous metastases from the stage III tumor. The patient subsequently received chemotherapy and radiation therapy followed by surgery of the rectosigmoid cancer.

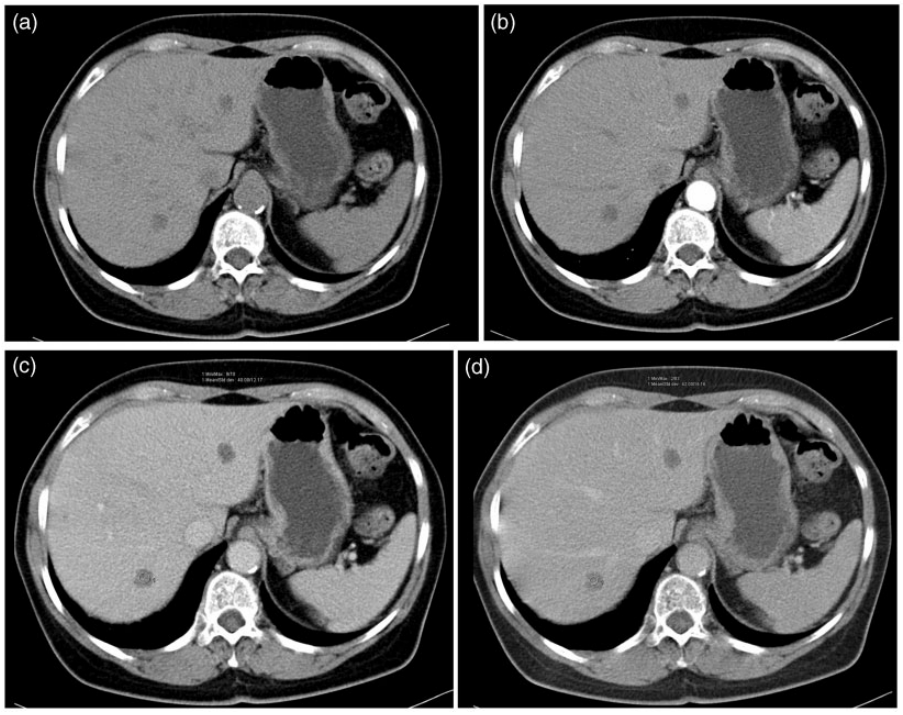

Fig. 1. A four phase CT scan through the liver. (a) Pre-contrast axial image showing two irregular low density (40 HU) lesions of 2 cm in segments II and VII. None of the lesions showed contrast enhancement. (b) Arterial phase. (c) Portovenous phase. (d) Late phase (5 min).

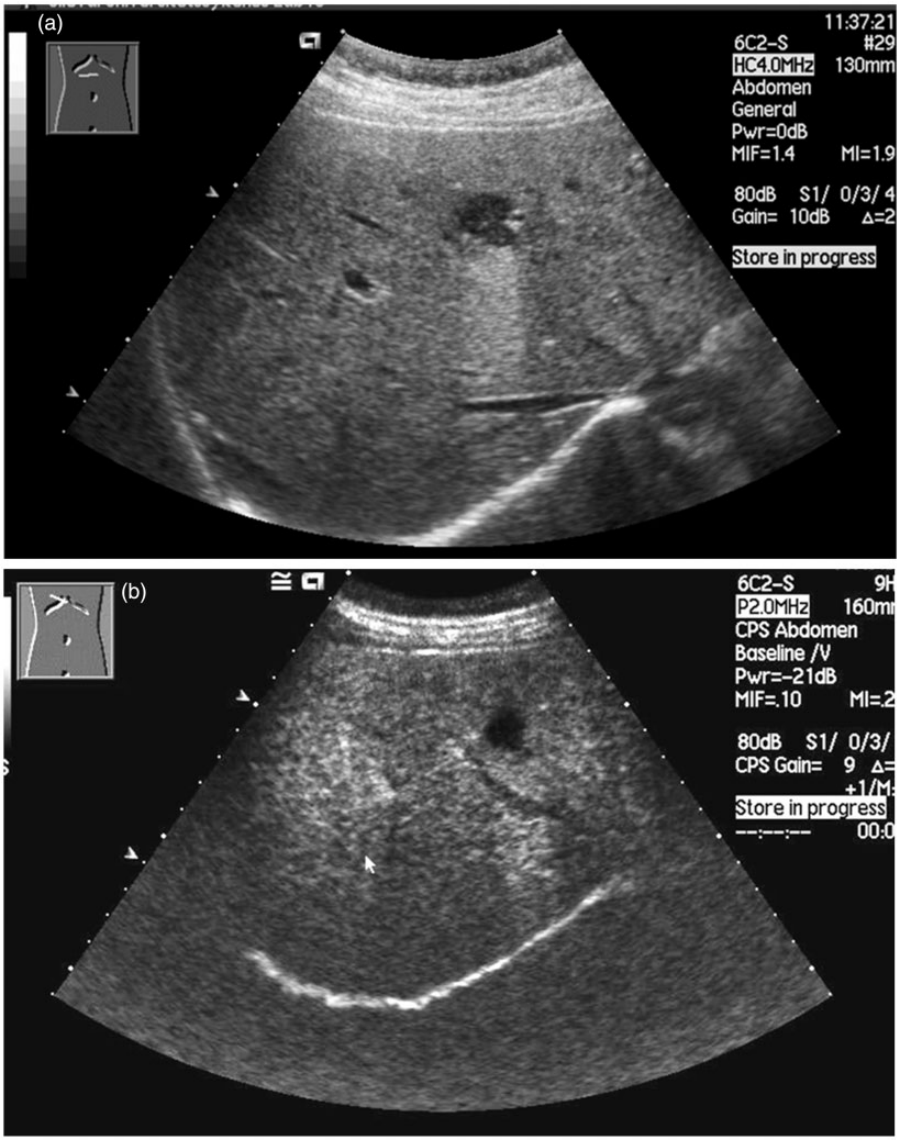

Fig. 2. Ultrasound of the liver with and without contrast. (a) Grayscale ultrasound showing a hypodense lesion in segment IV with an irregular border and a posterior echo-shadow indicating fluid content. (b) Contrast-enhanced ultrasound of the same lesion demonstrated no enhancement or washout in the arterial (not shown) or portal phase.

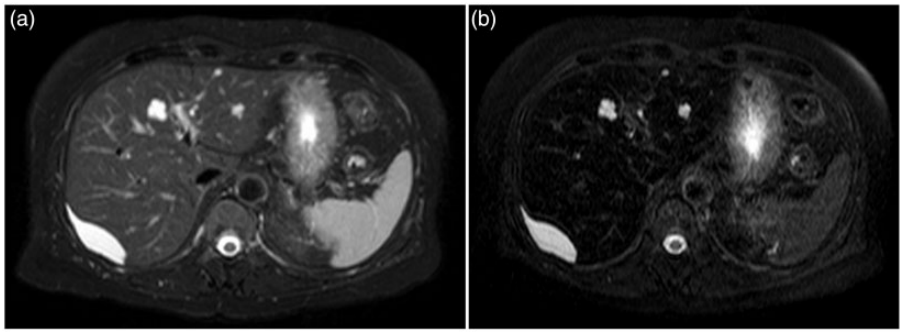

Fig. 3. MRI of the liver with and without SPIO. (a) Axial T2W sequence with fat suppression showing two hyper intense, lobulated cyst-like lesions in segments II and IV, and one 6 cm sub-capsular lens-shaped lesion in segment VII. (b) Following SPIO four additional sub-centimeter lesions were detected.

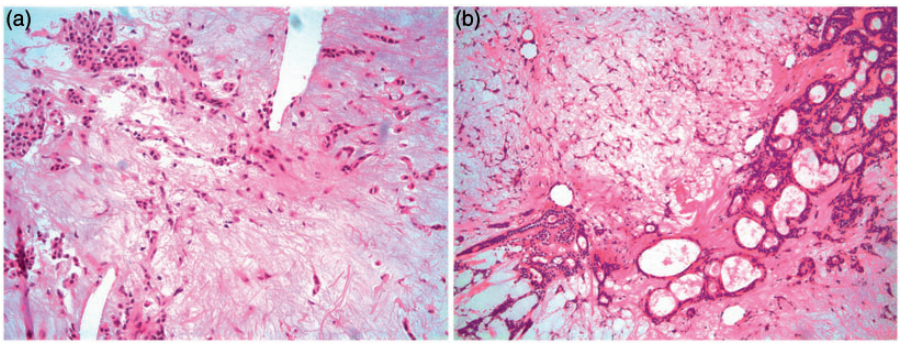

Fig. 4. Biopsy specimens (hematoxylin and eosin, 20× magnification). (a) The histopathology of the liver lesion in segment II showed the morphology of a so-called mixed tumor with a chondromyxoid stroma with irregular glandular structures and strands of relatively small uniform epithelial cells. (b) A 20-year-old stored specimen at our laboratory from a previous superficial parotidectomy of the same patient revealed the same histopathology pattern as the fresh liver biopsy with typical features of a pleomorphic adenoma.

A routine post-treatment evaluation 3 months later included a contrast-enhanced abdominal CT and MRI of the rectum. Both exams showed significant regression of the rectal tumor, while the liver lesions had not changed in size, numbers, or appearance. For a diagnostic re-assessment a percutaneous ultrasound-guided 18 G biopsy of one of the >1 cm lesions in liver segment II was obtained. The biopsy did not show features of a metastasis of colorectal cancer, but a chondromyxoid stroma and strands of small uniform epithelial cells resembling irregular glandular structures suggesting a pleomorphic adenoma. And indeed, retrieval of a previous specimen from a superficial parotidectomy performed 20 years earlier on this patient, and still stored at our laboratory, revealed the same histopathologic pattern as the biopsy of the focal liver lesion (Fig. 4).

Discussion

Initially, the lesions of our patient were interpreted as cystic metastases, on the basis of the patient's primary tumor, but since there were no morphologic changes following chemotherapy, it was reasonable to consider other (rare) differential diagnostic possibilities, and a biopsy was performed.

Most benign and metastatic liver lesions in cancer patients can be characterized with routine multimodality imaging with CT, US, and MRI, and a percutaneous biopsy is usually not required for a conclusive assessment and treatment decision (4–6). The finding of cyst-like hepatic lesions in a patient with a history of colorectal adenocarcinoma creates a diagnostic dilemma for both clinicians and radiologists. In our case none of the lesions showed contrast enhancement suggesting that common benign liver lesions like adenoma, Focal nodular hyperplasia (FNH), or hemangioma were unlikely. Correspondingly, hypo- and hypervascular metastases were also excluded. The patient had no symptoms or signs of infection; hence multiple hepatic abscesses were also considered unlikely. There was no previous history of liver disease and so cystic hepatocellular carcinoma and biliary cystic neoplasms as cystadenoma and cystadenocarinoma were less likely. There was no previous medical history of infection with tapeworm Echinococcus so hydaid cysts were considered very unlikely (4–6).

Most commonly cystic lesions in the liver represent simple liver cysts, and the incidence ranges up to 18–20% (4,7). Sonographically simple cysts present as anechoic lesions with a posterior acoustic enhancement, smooth borders and no septations. On CT the cysts were interpreted as water-attenuating lesions (<20 HU) with no visible wall and no contrast enhancement on CT. Cysts present with low signal intensity on T1W images and high signal intensity on T2W images and with no enhancement after contrast administration. Rarely, simple cysts appear as "complicated" cysts due to hemorrhage or inflammation, and are accordingly difficult to differentiate from cystic metastases, which remained as a possible differential diagnoses (5,6). Cystic metastases from a colorectal cancer are rare; a retrospective study from Sugawara found cystic colorectal metastases in 1.8% (8). On US they showed a heterogenous hypoechoic or anechoic pattern, and all lesions showed posterior echo enhancement, as in our case. On unenhanced CT, the lesions were seen as low-density masses with a predominantly homogeneous attenuation and the densities were in the range of 0–34 HU. The contours of the lesions were irregular in all but one case. On enhanced CT, the central portion of each tumor remained unenhanced and in two patients there was observed slight enhancement in the peripheral zones. One patient had lesions with septations, as in our case.

The imaging features of pleomorphic adenoma of the salivary gland is well described (9), and the previous

term, ''mixed tumor'' correlates well to both the histologic heterogeneity and the varying imaging patterns. Imaging findings may depend on the tumor size and the content of myxoid substance. It has been described that smaller adenomas appear more homogeneous and may enhance more strongly after contrast medium administration on both CT and MR (9). In our case both the CT, US, and MRI features showed no contrast enhancement reflecting the histopathology of a high myxoid (hypocellular) content of the lesions.

We have found only one case report of imaging of a benign pleomorphic adenoma with a metastasis to the liver (3). That patient presented with persistent abdominal pain and a prior history of a superficial parotidectomy 30 years earlier. US showed a large complex hyperechoic cyst with solid components. CT showed a large (11 × 16 cm) partly septated cyst. PET-CT showed a large hypermetabolic hepatic lesion with pathologic FDG uptake in both the solid and the septated components of the cyst. In this case the tumor was removed and microscopy revealed ''epithelial cells in abundant chondromyxoid stroma with no features of malignancy''. As in our case report, there was no evidence of local recurrence.

With regard to other abdominal organs, imaging of a BMPA to the kidney has been described (10). The BMPA represented as a solitary renal mass with hyperattenuation on CT and with peripheral calcifications. No cystic appearing components were observed. There was a history of local recurrence requiring multiple resections, 6 years prior. All in all, none of these two other reports of abdominal MBPA found morphological similarities on multimodality imaging with our case.

In conclusion, BMPA to the liver is extremely rare with no established characteristics on multimodality imaging. The radiologic appearance may mimic cystic metastases from colorectal cancer as illustrated in this report. In spite of aiming at a biopsy-less diagnostic workup, a percutaneous biopsy will be required in rare cases like this. Finally, a prior history of a pleomorphic adenoma may be crucial and suggestive of BMPA.

Conflict of interest

None declared.

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