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Intrahepatic Bile Duct Adenoma Masquerading as Hepatic Metastasis in a case of Carcinoma Rectum

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Abstract

Intrahepatic bile duct adenoma (BDA) is a rare benign tumor arising from the epithelium of the intrahepatic bile duct. In oncology, when a new onset hepatic nodule develops, it is often looked with suspicion for metastasis. However, many of these small liver nodules can be benign. Hence, it is important to obtain tissue for cytological/ histopathological diagnosis before labeling it as disease progression. We report imaging findings of bile duct adenoma, which was discovered during surveillance CT scan and was a new finding in a treated case of carcinoma anorectum. Since the new-onset, it was suspicious for metastasis on imaging. Ultrasound-guided FNAC was performed to confirm its etiology and the cytology report showed findings of bile duct adenoma.

Keywords: *Liver, Bile duct adenoma, metastasis, carcinoma rectum.*

Case History:

A 46-years-old female, a known case of adenocarcinoma of the rectum, received neoadjuvant chemo-radiotherapy and underwent posterior exenteration surgery. Histopathology report of the primary revealed a small residual focus of viable adenocarcinoma without nodal involvement. She received adjuvant chemotherapy and was on surveillance at a tertiary referral oncology center. Surveillance contrast-enhanced CT scan of abdomen and pelvis was obtained 9 months after the surgery on a 16-slice GE Discovery CT scan machine. Pre contrast plain images and post-contrast images in port venous phase (60 seconds after intravenous injection of iodinated contrast) and delayed / equilibrium phase (180 seconds after intravenous injection of iodinated contrast) were obtained. Arterial phase images were not obtained, as it was a routine surveillance CT scan. CT scan revealed a 14 mm-sized hypodense lesion in the right lobe of the liver, which was not evident on the previous pre-op CT. The liver lesion was hypoattenuating on pre-contrast plain images and post-contrast images in the port venous phase while showing hyperattenuating on delayed / equilibrium phase images. It was located at the periphery. (**Fig. 1**). Since it was a new onset lesion in a known case of carcinoma, it was suspicious for metastasis. The liver lesion seen on the CT scan did not show FDG uptake on the FDG PET-CT scan. (**Fig. 2**). Still, it was suspicious for metastasis since was new-onset. There is no evidence of metabolically active disease elsewhere in PET-CT. The case was discussed in the multidisciplinary tumor board and to know the exact pathology, whether benign or malignant, it was referred to us for ultrasound (USG) guided FNAC. When the patient was taken for ultrasound-guided FNAC, pre-procedural USG reveals an isoechoic lesion with a thin peripheral rim of hypoechogenicity (**Fig. 3**). Under all aseptic precautions & local anesthesia, USG guided FNAC was done with a 25 G spinal needle. Smears were prepared from the aspirate and sent for cytological examinations. On cytology, smears were cellular and showed groups of cuboidal epithelial cells with uniform nuclei, fine chromatin, and scanty cytoplasm. There was no atypia or mitoses. (**Fig. 4**) Hence cytological diagnosis was benign bile duct lesion, bile duct adenoma. The absence of atypia and mitoses ruled out metastasis and other neoplastic lesions like cholangiocarcinoma. In addition, the absence of dilated lumina and intraluminal bile ruled out Von Meyenberg Complexes. The patient was then put on routine surveillance.

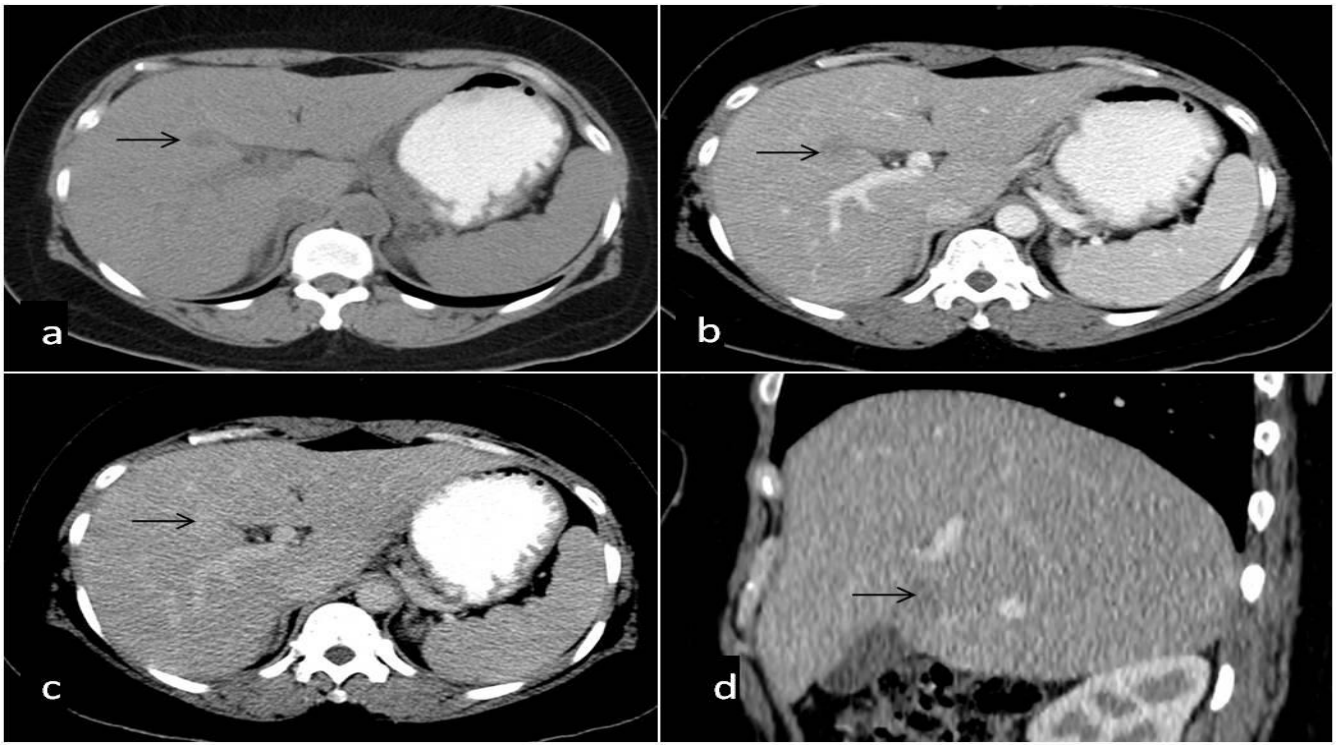

Figure 1: Axial CT scan non contrast (a) images and post contrast images in portovenous (b) and delayed phase (c) images show a hypodense nodule (arrows) which is hypoattenating to liver parenchyma on non contrast and post contrast portovenous phases, while is hyperattenuating on delayed phase images. (d) Sagittal reformatted image shows peripheral location of the lesion.

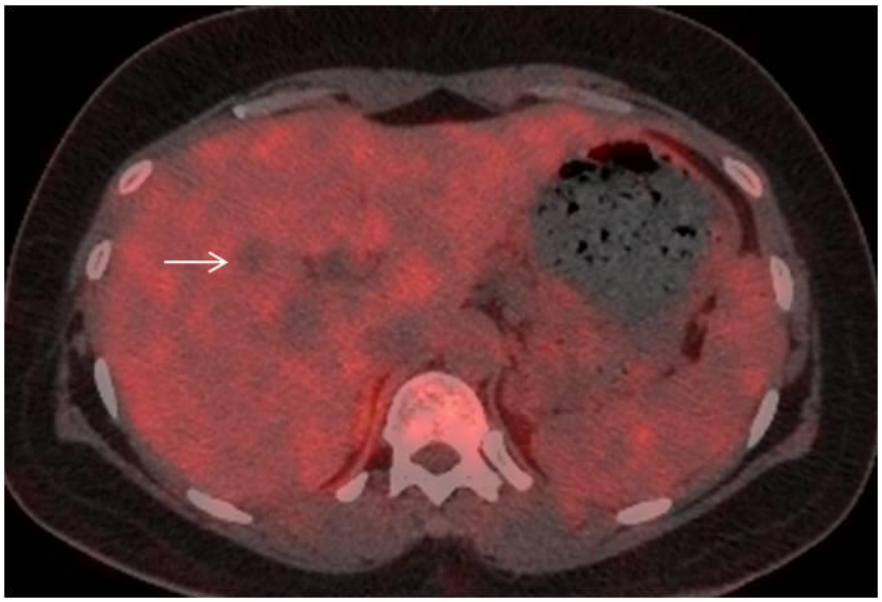

Figure 2: Axial fused images of PET CT scan shows no uptake in liver lesion (arrow)

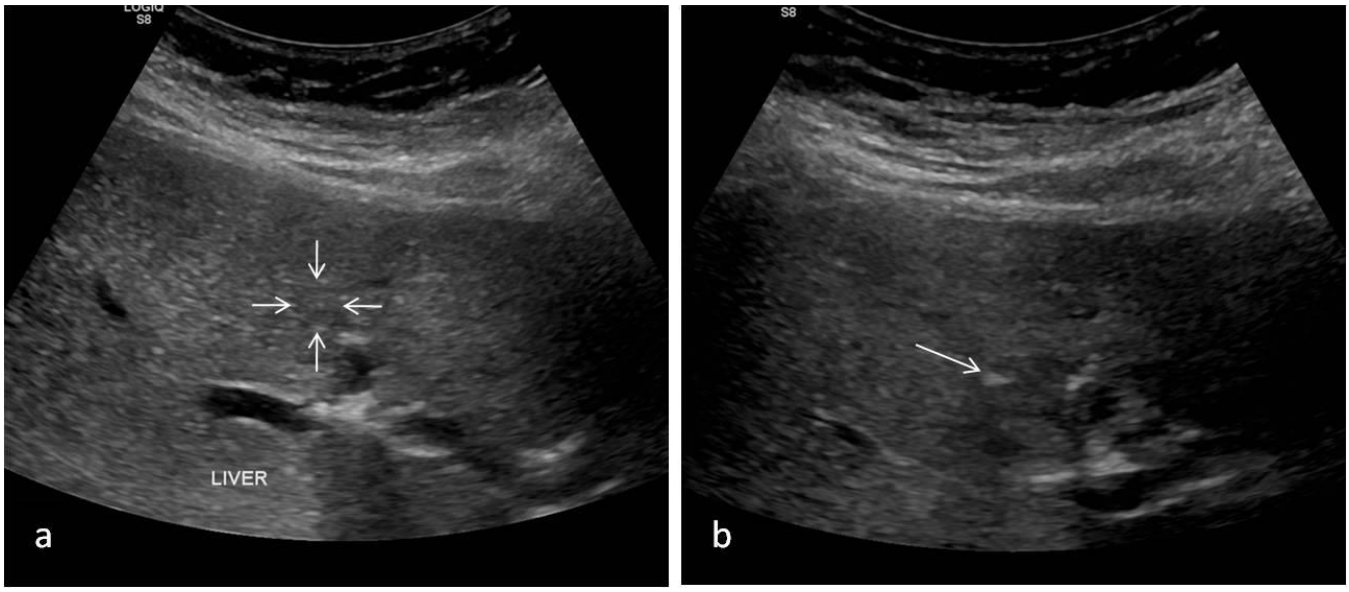

Figure 3: (a) Ultrasound images shows an isoechoic lesion with thin peripheral halo (marked with arrows). (b) USG guided FNAC was done. Arrow shows an echogenic tip of the needle in the liver lesion.

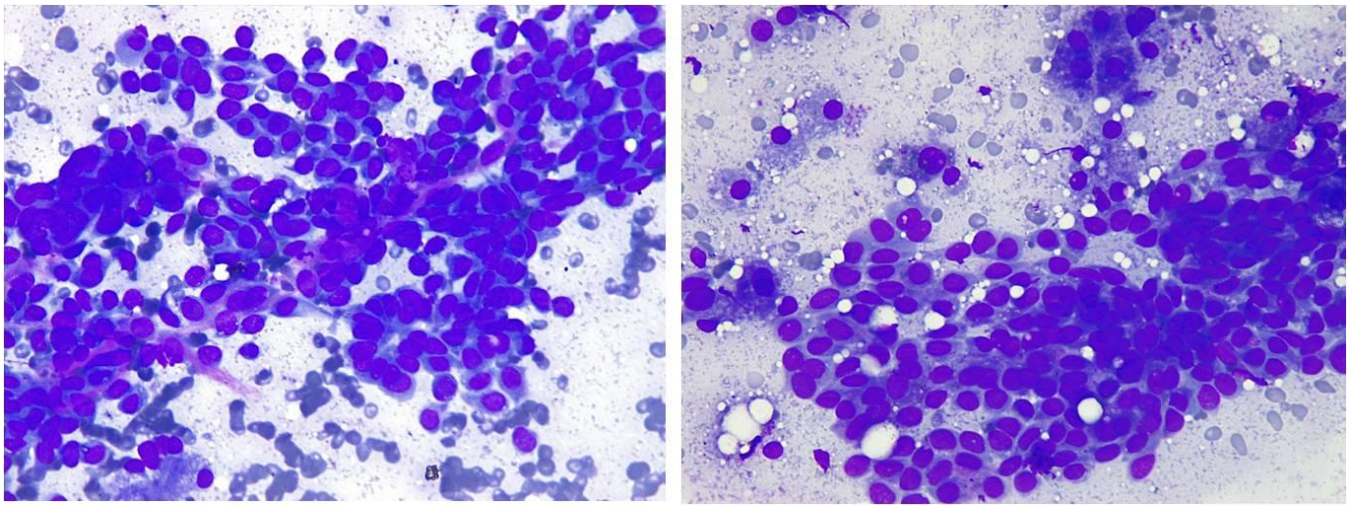

Figure 4: MGG stain 40x. It shows uniform cuboidal bile duct epithelial cells, with monomorphic nuclei, scanty cytoplasm. No atypia and no mitoses seen.

Discussion:

Bile duct adenoma is a rare benign tumor arising from the epithelial cells of the intrahepatic bile duct. **[**1,2,3**]** The largest studies to date by Allaire et al **[**1**]**, a study of 152 cases, found that most bile duct adenoma is asymptomatic and found incidentally during the intrabdominal surgery (103 case) or at autopsy (49). Bile duct adenoma constitutes around 1.3% of liver tumors. They are usually subcapsular

in location, range from 1 to 20 mm in size and were well-circumscribed but non encapsulated. The majority occurred in patients between 20 to 70 years, with a mean age of 55 years.[1]

Microscopically, BDA is seen as the proliferation of disorganized and mature peribiliary gland acini and ductules within a variable amount of connective tissue stroma with signs of chronic inflammation and collagenization. There is no atypia or mitosis, which helps in distinguishing them from cholangiocarcinoma. BDA can also be confused with bile duct hamartoma (Von Meyenberg Complex), but lack of dilated lumina and intraluminal bile helps differentiate BDA from hamartoma.[4]

On ultrasound, BDA appears as a hyperechoic nodule with or without a surrounding halo. They can be isoechoic to the liver and could not be identified or can be hypoechoic.[5,6] In our case, BDA was isoechoic to the liver parenchyma and shows a thin peripheral hypoechoic rim (peripheral halo).

On an unenhanced CT scan, BDA is hypodense. It can appear hyperdense which is due to calcifications within.[7] In arterial phase images, hyperenhancement is a common feature but may appear hypodense.[6] They are hyperattenuating relative to liver parenchyma on port venous and equilibrium phase images.[5,6,8] This delayed enhancement is likely due to fibrous stroma which is seen in BDA.[6,9] This enhancement may vary depending upon the amount of fibrous stroma and may appear hypodense on delayed phase images.[6] In our case, BDA was hypoattenuating to the hepatic parenchyma on the port venous phase and hyperattenuating on delayed phase images.

On MRI, BDA appears as hypointense relative to liver on T1-weighted images, hyperintense on T2-weighted images, and hyperintense on DWI. BDA also demonstrated characteristic features on dynamic enhanced MRI, i.e., hyperenhancement in portal venous and delayed phase images.[6,10,11] However, there are case reports which showed hypointense signal[7] and isointense signal [8] on T2 weighted images. MRI was not done in our case.

To our knowledge, there is no data in the literature about the role of FDG PET in intrahepatic BDA. Our case showed no uptake on FDG PET which was in favor of benign findings. However, some low-grade tumors, mucinous tumors and small lesions may not show FDG uptake [12] and given known primary malignancy with the new-onset liver lesion, it was reported as suspicious for metastasis.

In patients with known malignancy, definite characterization of small liver lesions into benign vs. malignant is crucial in determining the prognosis and treatment. Jones et al[13] reported in their study that liver lesions less than or equal to 15 mm were found in 17% of the cases and were benign in 51% of the 82% of patients with known malignancy. Schwartz et al[14] in their study reported that hepatic lesions less than or equal to 1 cm, deemed too small to characterize, are most often benign, but approximately 11.6 % of these lesions were malignant. Schwartz et al also reported in their study that when these too small to characterize liver lesions were followed, the average reported time for an increase in the size of these TSTC liver lesions was 13 months if malignant in etiology. Therefore, it is helpful to

obtain tissue for cytological/histopathological examination to obtain an accurate diagnosis and for better patient care.

"In conclusion, BDA, a rare primary liver tumor, have variable imaging characteristics ultrasound, but on dynamic contrast enhanced CT and MRI scans show arterial phase enhancement with progressive enhancement on delayed phase images, which are characteristics of BDA". However, some unusual imaging findings such as hypoattenuation on the equilibrium phase of CT scan are also reported in the literature. Being the rare tumor and possibility of unusual imaging findings, in a patient with known primary cancer, it is preferable to obtain tissue for cytology/histopathology to differentiate it from metastasis, which is more common than BDA.

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