CASE REPORT

Intrahepatic Bile Duct Adenoma Mimicking Hepatic Metastasis: Case Report and Review of the Literature

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We present a case of bile duct adenoma that mimicked hepatic metastasis from gastric cancer. The adenoma exhibited prolonged enhancement on dynamic computed tomography, hyperintensity on diffusion-weighted imaging, and diminished uptake of superparamagnetic iron oxide and gadolinium-ethoxybenzyl-diethylenetriamine pentaacetic acid on magnetic resonance imaging, resembling those of adenocarcinomas. Knowledge of the imaging findings of this rare entity may aid correct diagnosis.

Keywords: bile duct adenoma, diffusion-weighted imaging, gadolinium-ethoxybenzyl-diethylenetriamine pentaacetic acid, magnetic resonance imaging, superparamagnetic iron oxide

Introduction

Intrahepatic bile duct adenoma (BDA) is a rare benign epithelial tumor of the liver that is generally discovered incidentally during laparotomy or autopsy because of the lack of symptoms and small tumor size (one to 20 mm; mean, 5.8 mm).1 Histopathologically, BDA consists of a small aggregate of noncystic bile ductules associated with variable degrees of inflammation and fibrosis.1 Its etiology remains unclear. Previous reports have suggested that BDA is a reactive process to focal injury1 or a hamartoma that represents disorganized peribiliary glands.2 Computed tomography (CT) and magnetic resonance (MR) imaging are useful for detecting and diagnosing liver tumors. Detection of BDA will increase with the expanded use of and advances in cross-sectional imaging modalities, such as ultrasonography, CT, and MR imaging. However, only a few reports in the English literature describe imaging findings of BDA.3–5 To our knowledge, no imaging findings of BDA have been reported for diffusion-weighted (DWI) and superparamagnetic iron oxide (SPIO)-enhanced MR imaging.

We present a case of BDA in a patient with gastric cancer and describe findings on unenhanced and contrast-enhanced multidetector CT (MDCT) and MR imaging that included DWI and enhancement using SPIO and gadolinium-ethoxybenzyl-diethylenetriamine pentaacetic acid (Gd-EOB-DTPA). The benign tumor mimicked hepatic metastasis from gastric cancer.

Case Report

A 65-year-old woman with upper abdominal pain was referred to our hospital. Results of laboratory tests, including complete blood count, liver function tests, and tumor markers, were within normal limits. Esophagogastroduodenoscopy revealed an ulcerated lesion in the posterior wall of the body of the stomach. The gastric lesion was biopsied and determined pathologically to be adenocarcinoma. Preoperative ultrasonography revealed a hypoechoic nodule of 8-mm diameter near the hepatic capsule in the medial segment of the liver. MDCT was performed with a 16-MDCT scanner (Aquilion; Toshiba Medical Systems, Japan). The lesion was hypodense on unenhanced...
CT (Fig. 1A) and showed ring enhancement during the arterial phase (Fig. 1B) and isoattenuation compared to the surrounding liver parenchyma during the portal-venous phase (Fig. 1C) on dynamic CT. MR imaging was performed at 3 tesla (Magnetom Trio; Siemens AG, Erlangen, Germany). The lesion was hypointense on unenhanced fast low-angle shot T₁-weighted imaging (repetition time [TR], 133 ms; echo time [TE], 2.46 ms; flip angle [FA], 70°) (Fig. 2A) and isointense on respiratory-triggered fat-suppressed fast spin-echo T₂-weighted imaging (TR, 2693 ms; TE, 76 ms; FA, 180°; echo train length [ETL], 12) (Fig. 2B).

However, the BDA displayed clear hyperintensity on DWI (Fig. 2C) with an apparent diffusion coefficient (ADC) value of $1.10 \times 10^{-3}$ mm²/s. There was no signal drop in the lesion on SPIO-enhanced T₂*-weighted imaging (TR, 205 ms; TE, 9.84 ms; FA, 60°) (Fig. 2D). On Gd-EOB-DTPA-enhanced MR imaging using 3-dimensional fat-suppressed T₁-weighted gradient-echo volumetric interpolated breath-hold examination (TR, 3.4 ms; TE, 1.24 ms; FA, 10°), the lesion showed peripheral enhancement during the hepatic arterial phase and relative hypointensity compared to the surrounding liver parenchyma during the portal and transitional phases but distinct hypointensity during the hepatobiliary phase (Fig. 2E). These findings suggested hepatic metastasis from gastric cancer. The patient received systemic chemotherapy using combined anticancer drugs tegafur, gimeracil, and oteracil potassium. After 5 months, the gastric lesion decreased in size, but the hepatic lesion did not change. The patient underwent total gastrectomy and wedge resection of the hepatic lesion. Grossly, the resected specimen of the liver had a well defined and nonencapsulated yellowish white solid lesion (Fig. 3A). Microscopic examination revealed proliferation of epithelial cells of the bile ductules in the peripheral area (Fig. 3B), which cor-
Fig. 2. (A) Fast low-angle shot T1-weighted magnetic resonance (MR) imaging shows a homogeneous hypointense lesion (arrow). (B) On fast spin-echo T2-weighted MR imaging, the lesion shows homogeneous isointensity (arrow). (C) Diffusion-weighted MR imaging with a b-value of 1000 s/mm² shows a homogeneous hyperintense lesion (arrow). (D) On superparamagnetic iron oxide (SPIO)-enhanced T1*-weighted MR imaging, the lesion appears hyperintense relative to the surrounding liver parenchyma. (E) During the hepatobiliary phase of gadolinium-ethoxybenzyl-diethylentriamine pentaacetic acid (Gd-EOB-DTPA)-enhanced MR imaging, the lesion exhibits distinct hypointensity.

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responded to the area that showed ring enhancement during the arterial phase on dynamic CT. The central area that showed delayed enhancement on dynamic CT corresponded to dense fibrous stroma (Fig. 3C). Immunohistochemical examination demonstrated positive staining for cytokeratin 7 and 19. Pathological diagnosis of the liver lesion was BDA and of the gastric lesion, poorly differentiated adenocarcinoma.

Discussion

Only 3 reports in the English literature have included radiological findings of 5 BDAs.3–5 Table summarizes the imaging findings of those cases and ours. Five of the 6 BDAs (including our case) showed delayed or prolonged enhancement during the late phase in dynamic CT. This enhancement pattern may be attributable to abundant fibrous stroma within the tumor and resembles the pattern of adenocarcinomas with abundant fibrous stroma, such as cholangiocarcinoma and metastasis from cancer of the gastrointestinal tract cancer.6 In our case, we observed delayed enhancement in the central area with dense fibrous stroma.

On T2-weighted MR imaging, most metastatic adenocarcinomas are hyperintense, but some adenocarcinomas with dense fibrosis exhibit iso- or hypointensity. On T2-weighted imaging, four of the 5 previously reported BDAs exhibited hyperinten-
Fig. 3. (A) Gross specimen shows a nonencapsulated, whitish, subcapsular lesion. (B) Microscopic findings show proliferation of epithelial cells of the bile ductules in the peripheral area (hematoxylin-eosin, × 200). (C) Microscopic findings show bile ductules with dense fibrous stroma in the central area (hematoxylin-eosin, × 200).

sity; the fifth exhibited hypointensity. Our case exhibited isointensity on T2-weighted imaging, which could be attributed to abundant fibrous stroma within the lesion.

Our case of BDA exhibited hyperintensity on DWI. DWI reflects changes in tissue water mobility, provides different tissue contrast from that of conventional T1- and T2-weighted imaging, and has been used to detect and diagnose liver tumors.\(^7,8\) Parikh and colleagues\(^8\) reported the superiority of DWI to standard T2-weighted imaging for detecting focal hepatic lesions. We clearly visualized the BDA in our case on DWI, but it was not clear on T2-weighted imaging. The ADC value was 1.10 × 10\(^{-3}\) mm\(^2\)/s in the present case. The ADC values of focal nodular hyperplasias and liver cell adenomas have been reported to be similar to those of hepatocellular carcinomas (HCCs) and metastases.\(^7\) Thus, it might be difficult to differentiate BDA from hepatic metastasis on the basis of DWI.

SPIO is a tissue-specific MR imaging agent taken up by Kupffer cells in the liver. Gd-EOB-DTPA is a new hepatocyte-specific contrast agent taken up by hepatocytes during the hepatobiliary phase. SPIO- and Gd-EOB-DTPA-enhanced MR images are used to improve detection and characterization of focal hepatic lesions in clinical studies.\(^9,10\) On SPIO-enhanced MR imaging, we visualized the present BDA as an area of diminished SPIO uptake, such as that observed in hepatic metastatic lesions. During the hepatobiliary phase of Gd-EOB-DTPA-enhanced MR imaging, our case and one previously reported case\(^9\) demonstrated distinct hypointensity. As with a hepatic metastatic lesion, a BDA lacks Kupffer cells and normal hepatocytes, which results in diminution of SPIO and Gd-EOB-DTPA uptake and difficulty in differentiating BDA from malignant liver tumors.

This is the first report on DWI and SPIO-enhanced MR imaging findings of BDA. DWI is useful for detection and characterization of hepatic tumors.\(^7\) SPIO- and Gd-EOB-DTPA-enhanced MR imaging can improve sensitivity in detecting hepatic metastatic lesions.\(^9,10\) However, we found it difficult to differentiate BDA from a hepatic metastatic lesion from gastric cancer, even with the combination of dynamic CT, DWI, and SPIO- and Gd-EOB-DTPA-enhanced MR imaging. Though rare, BDA should be included in the differential diagnosis of small hypervascular and prolonged-enhancing tumors in the periphery of the liver.
### Table. Summary of reported bile duct adenomas

<table>
<thead>
<tr>
<th>Reporter</th>
<th>Age, Gender, Hepatitis</th>
<th>Location</th>
<th>Unenhanced computed tomography</th>
<th>Dynamic study</th>
<th>EOB-MRI hepatobiliary phase</th>
<th>SPIO T₂*WI</th>
<th>ADC (x 10⁻³ mm²/s)</th>
<th>Macroscopic findings</th>
<th>Dynamic study</th>
<th>EOB-MRI hepatobiliary phase</th>
<th>SPIO T₂*WI</th>
<th>ADC (x 10⁻³ mm²/s)</th>
<th>Macroscopic findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tajima T et al. (3)</td>
<td>75/m/B</td>
<td>Posterior segment of the right lobe, surface</td>
<td>Hypoechoic</td>
<td>Hypodense on early and delayed phase</td>
<td>Good enhancement during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>NA</td>
<td>Round/yellowish white mass/5 mm</td>
<td>Hypo, hyper, NA</td>
<td>Hypodense during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>NA</td>
<td>Round/yellowish white mass/4 mm</td>
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<tr>
<td>66/f/C</td>
<td>Lateral segment of the left lobe, subcapsular</td>
<td>Not detected</td>
<td>Hypodense</td>
<td>Hypodense on early and delayed phase</td>
<td>Good enhancement during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>NA</td>
<td>Round/yellowish white mass/10 mm</td>
<td>Hypo, hyper, NA</td>
<td>Hypodense during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>NA</td>
<td>Round/yellowish white mass/20 mm</td>
</tr>
<tr>
<td>Maeda E et al. (4)</td>
<td>78/f/none</td>
<td>Posterior segment of the right lobe, surface</td>
<td>Hypoechoic</td>
<td>Hypodense</td>
<td>Periferal enhancement during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/20 mm</td>
<td>Hypo, iso, hyper</td>
<td>Hypodense during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/18 mm</td>
</tr>
<tr>
<td>59/m/B</td>
<td>Anterior segment of the right lobe, subcapsular</td>
<td>Not detected</td>
<td>Hypodense</td>
<td>Hypodense</td>
<td>Periferal enhancement during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/20 mm</td>
<td>Hypo, iso, hyper</td>
<td>Hypodense during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/18 mm</td>
</tr>
<tr>
<td>65/m/none</td>
<td>Medial segment of the left lobe, subcapsular</td>
<td>Hypoechoic</td>
<td>Hypodense</td>
<td>Hypodense</td>
<td>Periferal enhancement during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/20 mm</td>
<td>Hypo, iso, hyper</td>
<td>Hypodense during arterial phase and prolonged enhancement</td>
<td>NA</td>
<td>1.1 Hyperintense</td>
<td>Round/yellowish white mass/18 mm</td>
</tr>
<tr>
<td>Present</td>
<td>Brown, yellow</td>
<td>ADC, apparent diʃusion coeʃcient; DWI, diʃusion weighted image; EOB-MRI, gadolinium-ethoxybenzyl-diethylenetriamine pentacetic acid magnetic resonance imaging; NA, not available; SPIO, superparamagnetic iron oxide; T₁WI, T₁-weighted image; T₂WI, T₂-weighted image; T₂<em>WI, T₂</em>-weighted image; US, ultrasonography</td>
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References