# **MRI Findings in Biliary Cystadenoma** Güray Can<sup>1</sup>, Sedat Alpaslan Tuncel<sup>2</sup>, Hakan Gençhallaç<sup>2</sup>, Fulya Öz Puyan<sup>3</sup>, Cem İbiş<sup>4</sup>

Biliary cystadenoma is a rare cystic hepatic neoplasm. We herein present a 44 year-old female patient with biliary cystadenoma focusing on difficulties in diagnosis because of lack of specific findings with imaging, and the need for surgery. As biliary cystadenomas have high recurrence risks and malignant transformation potential, careful follow-up should not be underestimated. Proper surgical resection of the lesion may be considered the most suitable diagnostic and therapeutic procedure, and prognosis is generally excellent after removal of the tumor.

Key words: Cystadenoma, biliary tract, MRI

## Biliyer Kistadenomda MR Bulguları

## ÖZET

Biliyer kistadenom karaciğerin nadir görülen bir kistik neoplazmıdır. Kistadenom ifadesi benign süreci düşündürmekle birlikte, bu lezyonlar malign transformasyon potansiyeline sahip olup, cerrahi sonrası da rekürrens riski bulunması nedeniyle dikkatli takip gerektirmektedir. Lezyonun uygun radyolojik görüntülemesi sonrasında yapılacak cerrahi tedavi ile prognoz genellikle mükemmeldir. Biz radyolojik görüntülemenin spesifik tanıya yardımcı olduğu ve cerrahi tedavi uygulanmış, 44 yaşında biliyer kistadenomlu kadın olguyu sunuyoruz.

Anahtar kelimeler: Kistadenom, safra kanalı, MRG

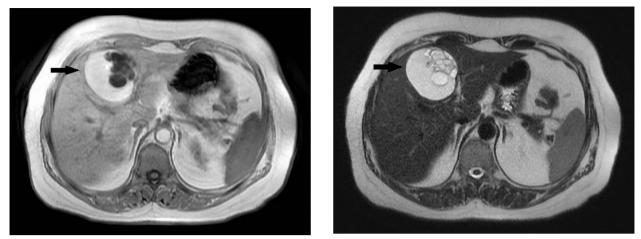
# INTRODUCTION

Biliary cystadenoma is a rare cystic hepatic neoplasm (1). Though generally accepted as a recurrent but benign lesion, it has a potential for malignant transformation. Tumor originates from intrahepatic bile ducts and lined by mucin-secreting epithelium. It appears as a unilocular or multilocular hepatic mass, encircled by a thick fibrous capsule. Internal septations and intraluminal projections may be present within the tumor. Biliary cystadenomas are often found incidentally, during a routine checkup, with certain imaging modalities such as ultrasound (US), magnetic resonance imaging (MRI) or computed tomography scans (CT). Patients are usually asymptomatic or present with nonspecific symptoms such as blunt abdominal pain or abdominal fullness due to local compression and mass effect. Symptoms may vary according to the size and location of the tumor; while a tumor in the right upper quadrant can cause

Trakya University Faculty of Medicine, Departments of Gastroenterology' Edirne, Radiology<sup>2</sup>, Pathology<sup>3</sup> and General Surgery<sup>4</sup>, Edirne, Turkey Received: 06.01.2014, Accepted: 01.06.2014 abdominal tenderness, a large tumor may present as hepatomegaly. Both biliary cystadenoma and its malignant form cystadenocarcinoma are slow growing tumors, making the differential diagnosis difficult, since neither presentation nor findings of examination or imaging is specific for either entity. Certain cystic lesions of the liver, including hydatid cysts are radiologically similar, almost indistinguishable from cystadenomas and cystadenocarcinomas. Accurate diagnosis depends on pathological evaluation. As the tumor has high recurrence rates and malignant potential, surgical resection is inevitable. The mortality and morbidity rates of hepatic cystadenomas are reported to be low after curative resection.

We herein present a case with cystadenoma focusing on difficulties in diagnosis because of lack of specific findings with imaging, and the need for surgery.

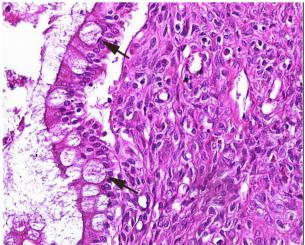
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**Figure 1.** Pathologically proven hepatobiliary cystadenoma. A single, cystic mass with papillary infoldings; a mostly hypointense but partially hyperintense on axial T1-weighted images (hyperintense area is compatible with high protein content in the cystic mass), b hyperintense on axial T2-weighted images (arrows).

# CASE

A 44 year-old female patient presented with blunt epigastric pain for almost a month. She did not have a remarkable medical history; she did not have any chronic disease, and was not taking regular medications. Her physical examination was normal except for mild epigastric tenderness. Whole blood count and liver transaminase levels were normal. Serological tests for hepatitis B, hepatitis C and echinococcus granulosus were



**Figure 2.** High power view of the cyst epithelium shows benign ciliated mucinous cells and scattered goblet cells (arrows) (H&E x 200).

negative. A cyst, 68x59 mm in size, with lobulated contours and multiple thin septations located in the left lobe of the liver was observed with US. CT demonstrated minimal contrast enhancement within the septations and the wall of the cyst. MRI (1.5 T MR unit/Signa; GE Medical System, Milwaukee, WI) showed a single cystic mass with papillary infoldings, mostly hypointense and partially hyperintense on T1-weighted (hyperintense area was compatible with high protein content in the cystic mass), and hyperintense on T2 weighted sequences (Figure 1a,b). The mass was surgically resected. It was a 18x13x4 cm sized, 510 gram weighted material containing multilocular, 6 cm sized cystic mass. The cyst was filled with mucinous and yellow colored material. Microscopically, the cyst was lined by a single layer of mucin-secreting columnar epithelial cells with bland, basally located nuclei. Goblet cells were intermingled between mucinous epithelial cells (Figure 2). A fibrotic stroma resembling ovarian stroma surrounded the epithelial cells. Histological evaluation confirmed the diagnosis of hepatobiliary cystadenoma (endocervical and focal intestinal type). The patient was discharged from the hospital without any complications. Informed consent was obtained from the patient.

# DISCUSSION

Hepatobiliary cystadenoma is a rare, benign cystic tumor of the liver. It usually occurs in middle-aged women (1). Though generally considered as a benign tumor, it has a potential for malignant transformation, and may turn into cystadenocarcinomas (2). Etiology of both entities remain unclear, although cystadenoma is considered a congenital condition developing from aberrant hamartomatous bile ducts, cystadenocarcinomas develop as a complication of antecedent cystadenomas (3,4).

Preoperative diagnosis of cystadenomas is guite challenging (1). Laboratory parameters are generally useless, mildly elevated liver transaminases may be present. Levels of tumor markers such as carcinoembryonic antigen and carbohydrate antigen 19-9 (CA 19-9) may be elevated in biliary cystic tumors though they are not sensitive or specific for cystadenomas, cystadenocarcinomas or any other cystic lesions of the liver (1,5-7). Cytological examination of the cystic fluid is not considered as a reliable method for diagnosis (8.9). Like other cases in the literature, all laboratory parameters were within normal limits in our patient. Imaging is the most widely used technique in the differential diagnosis of liver lesions. Ultrasound, as well as CT provides most of the data necessary to establish the diagnosis. Biliary cystic tumors are determined as their being uni- or multilocular and also the presence of intracystic projections (10,11). The diagnosis of biliary cystic tumor is likely if uni- or multilocular cystic lesions and papillary infoldings are present in US or CT imaging. Contrast-enhanced CT reveals the cystic wall and papillary infoldings, while US provides an image of a hypoechoic cystic mass with echogenic septations or papillary infoldings (10-12). The US appearances of both biliary cystadenoma and cystadenocarcinoma are guite similar; multiple septations with thick, single and hyperechoic outer wall. Vascular flow within the mass may also be determined (1). These tumors are usually described as well-demarcated lesions with contrast-enhanced walls and septae. Coarse calcifications along the wall or septa in a multilocular cystic mass may suggest a cystadenocarcinoma (10). On both T1- and T2-weighted MRI sequences, cysts have variable signal intensity, depending on the protein content and the presence or absence of hemorrhage (1). In our case, although MRI revealed partial hyperintensity on T1weighted sequences, analysis of the cystic fluid showed a high protein content rather than a hemorrhagic fluid. Likewise, CT revealed minimal contrast enhancement of the thin septations and cyst wall, and MRI demonstrated a single cystic mass with papillary infoldings; mostly hypointense and partially hyperintense on T1-weighted, and hyperintense on T2-weighted sequences. Although septal nodularity is interpreted as an indicator of cystadenocarcinomas, imaging alone is not sufficient in differential diagnosis (10).

It is essential to identify cystadenomas and cystadenocarcinomas as well as to distinguish both entities since management of each entity is different from that of any other hepatic cystic lesions. Generally accepted treatment modalities include aspiration, drainage and marsupialization. Because of their recurrence and malignant transformation potential, careful follow-up should not be underestimated (13). Proper surgical resection of the lesion is a decent diagnostic and therapeutic procedure, and prognosis is generally excellent after removal of the tumor.

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