

Hepatobiliary Cystadenoma with Mesenchymal Stroma Mistaken for Common Bile Duct Stone

A. H. Mohammad Alizadeh,
F. Moradi Moghaddam, A. Hekmatdoost,
H. Kalbasi

Abstract

A 65-year-old female presenting with hepatobiliary cystadenoma (HC) comprised of multiple cysts in the liver and a non-specific epigastric pain is reported herein. Ultrasound study demonstrated dilated common bile duct (CBD) and intrahepatic bile ducts, plus two spherical stone-like bodies filling defects in the proximal CBD. Sphincterotomy and basket performed for the patient were not successful. Upon surgery, two cystic masses were found in CBD and were resected. Histopathological examinations showed hepatobiliary cystadenoma with mesenchymal stroma.

Iran J Med Sci 2004; 145-147.

Keywords • Hepatobiliary • cystadenoma • hepatobiliary stone • biliary cystadenoma

Introduction

Biliary cystadenoma is a rare mass occurring primarily in women in the fifth decade of their life, while extrahepatic bile duct involvement with multiple cysts is an uncommon occurrence.¹⁻³ These tumors comprise less than 5% of all intrahepatic cysts of biliary origin.⁴ Clinical presentation is usually mild and atypical,⁵ and a definitive diagnosis can be made by endoscopic retrograde cholangiopancreatography (ERCP) or spiral computed tomography (CT).⁶ Herein, we describe a case of cystadenoma with mesenchymal stroma, which was misdiagnosed as a common bile duct (CBD) stone.

Case Report

In December 2003, a 65-year-old woman was admitted to our clinic with a two-year history of abdominal pain. She presented with epigastric and right upper quadrant (RUQ) mild pain with no radiation. She had undergone appendectomy and cholecystectomy when 35 and 40 years old respectively.

Physical examination was unremarkable, except for the presence of mild epigastric tenderness. Laboratory data, on admission, revealed only a mild anemia (Hb=11.09 mg/dl). Other biochemical and liver function tests were normal. Abdominal ultrasound showed dilatation of the proximal part of the CBD. Furthermore, ERCP demonstrated dilatation of common, extra, and intrahepatic bile ducts associated with two stone-like bodies measuring 10×12 and 9×10 mm filling defects in the proximal part of CBD.

According to the ERCP findings and considering the high prevalence of CBD stone in our region, sphincterotomy and basket were performed with failure. The patient was then referred to the surgical ward. Abdominal computed tomography

Research Center for Gastroenterology and Liver Disease,
Shaheed Beheshti University of Medical Sciences,
Tehran, Iran

Correspondence:
A Hekmatdoost MD
Research Center for Gastroenterology and Liver Disease
Shaheed Beheshti University of Medical Sciences
Tehran, Iran
Tel:+98-21-2418871
Fax:+98-21-2402639
E-mail: 89010@dedicated.irimc.org



Fig 1: CT scan findings shows multiple cystic lesions.

The CBD performed before the operation revealed dilatation of associated with multiple cystic lesions in the liver and a cyst in the proximal CBD (Fig 1). Thus, the patient underwent surgical operation for the resection of cystic lesions. On operation, two cystic lesions (one in the proximal CBD (10×12mm) and the other in the hilum of the liver (9×10mm) was found and resected. Pathological examination revealed a mucin-producing cyst underlined by a dense layer of mesenchymal stroma cells, which was compatible with hepatobiliary cystadenoma with mesenchymal stroma (Fig 2). Six months after surgery, no evidence of recurrence of the cyst was found and the patient is doing well.

Discussion

HC are rare but because of their potential malignancy deserve correct diagnosis. They are usually solitary lesions accompanied by multilocular cysts in the liver. Despite available diagnostic modalities they are difficult to differentiate from cystadenocarcinoma. There are numerous reports concerning misdiagnosed HC as hydatid cysts,⁷⁻⁹ with a single report on hepatobiliary cystadenoma resembling biliary smooth muscle neoplasm.⁹

Our patient was presented with nonspecific symptoms. Ultrasound could not detect the cysts and ERCP findings were in favor of CBD stone. Sphincterotomy and basket were performed to extract the putative stone but due to misdiagnosis, they were not successful. Finally, preoperative computed tomography revealed the cysts.

The first case of hepatobiliary cystadenoma was reported in 2001.¹ In this regard, this is the second of such a report involving multiple simple liver cysts. However, hydatid cyst, or hepatobiliary smooth muscle neo-

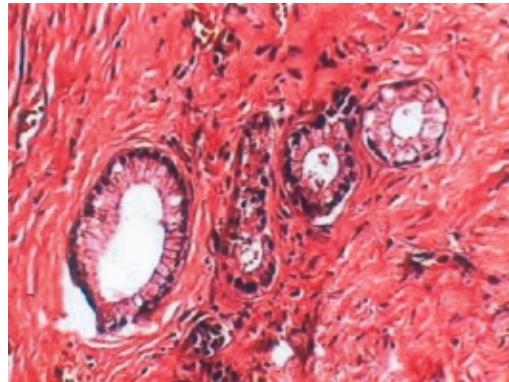


Fig 2: Microscopic view of resected cyst indicates a mucin producing cyst underlined by a denser layer of mesenchymal stroma cells.

plasm and common bile duct stone should be considered for differential diagnosis of cystadenomatosis.

References:

- 1 Hara H, Morita S, Sako S, Dohi T, Iwamoto M, Inoue H, Tanigawa N. Hepatobiliary cystadenoma combined with multiple liver cysts: report of a case. *Surg Today*. 2001; **31**: 651-4.
- 2 Devaney K, Goodman ZD, Ishak KG. Hepatobiliary cystadenoma and cystadenocarcinoma: a light microscopic and immunohistochemical study of 70 patients. *Am J Surg Pathol* 1994; **18**:1078-91.
- 3 Ishak KG, Willis GW, Cummins SD, Bullock AA. Biliary cystadenoma and cystadenocarcinoma: report of 14 cases and review of the literature. *Cancer* 1977; **38**: 322-38.
- 4 Takayasu K, Muramatsu Y, Moriyama N, et al. Imaging diagnosis of bile duct cystadenocarcinoma. *Cancer* 1988; **61**: 941-46.
- 5 Organ B, Petrak J: Biliary cystadenoma. *South Med J* 1984; **77**: 262-65.
- 6 Van Steenbergen W, Ponette E, Marchal G, et al. Cystadenoma of the common bile duct demonstrated by endoscopic retrograde cholangiography: an uncommon cause of extrahepatic obstruction. *Am J Gastroenterol* 1984; **79**: 466-70.
- 7 De Backer AI, De Schepper AM, Michielsen PP, et al. Hepatobiliary cystadenoma with mesenchymal stroma mimicking hydatid cyst: Report of a case. *Acta Gastroenterol Belg*. 2000; **63**: 317-20.
- 8 Kapoor R, Calton N, Verghese M. Cystadenoma with mesenchymal stroma mistaken for hepatic hydatid cyst. *Indian J Gastroenterol*. 1997; **16**: 73-4.

Hepatobiliary cystadenoma mistaken for stone

- 9 Gadzijev E, Dragan S, Verica FM, Jana G. Hepatobiliary cystadenoma protruding into the common bile duct, mimicking compli-

cated hydatid cyst of the liver: Report of a case. *Hepatogastroenterology*. 1995; **42**: 1008-10.