CASE REPORT

TJG
The Turkish Journal of Gastroenterology

A case of biliary cystadenocarcinoma with malignant seeding of the tract after percutaneous catheter drainage

Ma YUEFENG[1], Fu WEILI[1], Lan JING[1], Tan WENXIANG[1], Liu TIANQING[2], Wang LIMING[3]

Departments of [1]Hepatobiliary Surgery and [2]Pathology Dalian Friendship Hospital of Dalian Medical University, Dalian Liaoning, China

[3]Organ Transplantation Center, The Second Affiliated Hospital of Dalian Medical University, Dalian, Liaoning, China

Biliary cystadenocarcinoma is rare tumor that originates from the hepatobiliary epithelium, and its clinical diagnosis is difficult during the preoperative course. A 65-year-old woman with biliary cystadenocarcinoma was misdiagnosed as hepatic abscess and underwent ultrasonography-guided percutaneous catheter drainage. Ten months later, the patient was re-admitted to our department with a mucin-producing cauliflower-like mass measuring 10x10x5 cm^3 at the site of puncture. Tumor seeding through the percutaneous catheter drainage tract was diagnosed. Complete resection of the primary and metastatic tumor with partial abdominal wall tissue was performed. No local recurrence could be found after a follow-up of more than two years.

Key words: Cystadenocarcinoma, liver abscess, percutaneous catheter drainage

Perkütan kateter drenajı ertesinde traktusta malign yayılım görülen biliyer kistadenokarsinom vakası

Safra yolu kistadenokarsinomu hepatobiliyer epitelden köken alan nadir görülen bir tümördür ve preoperatif tanısı zordur. Karaciğer absesi tanısı yanlışlıkla konulmuş olan 65 yaşında kadın hastaya ultrason eşliğinde perkütan drenaj uygulanmıştır. On ay sonra hasta kliniğimize ponksiyon yerinde gelişen 10x10x5 cm boyutlarında müsin üreten karnıbahar şeklinde kitle ile başvurmuştur. Perkütan girişim hattında tümör yayılımı tanısı konulmuştur. Primer tümör ile beraber metastazı bir miktar abdomen duvarı ile beraber rezeke edilmiştir ve 2 yıllık takibin sonunda lokal rekürrens tespit edilmemiştir.

Anahtar kelimeler: Kistadenomarsinom, karaciğer absesi, perkütan kateter drenajı, malign yayılım

INTRODUCTION

Biliary cystadenocarcinoma (BCAC) is a rare tumor that arises in the liver or, less frequently, in the extrahepatic biliary system. Because of insufficient recognition of this disease and its indistinctive clinical presentation, it is often misdiagnosed (1) and even improperly treated (2).

Recently, we encountered a patient with BCAC who, after having undergone percutaneous catheter drainage (PCD) on the basis of an initial misdiagnosis of liver abscess, developed cancer seeding along the PCD tract.

CASE REPORT

In August 2008, a 65-year-old woman was referred to our institute for rigor, fever and abdominal pain in the right upper quadrant that had continued for several weeks; her physical examination revealed epigastric tenderness. Laboratory examination was

Address for correspondence: Wang LIMING
The Second Affiliated Hospital of Dalian Medical University, Organ Transplantation Center, Dalian/Liaoning, China
E-mail: wanglimingmd@hotmail.com

Manuscript received: 09.03.2012 **Accepted:** 17.04.2012

Turk J Gastroenterol 2012; 23 (6): 795-798
doi: 10.4318/tjg.2012.0644

unremarkable except for elevation in serum aminotransferases and leukocytes. An abdominal computed tomography (CT) scan showed a well-defined solitary and unilocular cyst in segments II and III of the liver. After an initial diagnosis of hepatic abscess, ultrasonography-guided PCD was performed, and a large amount of thick mucinous material was aspirated; the catheter was withdrawn 15 days later. Aspiration cytology and histological examination were performed three times; no malignant cells were found. With the suspicion of biliary cystadenoma or cystadenocarcinoma, exploratory laparotomy was recommended; however, the patient refused the procedure. The patient was asked to attend follow-up regularly, but failed to follow this advice.

Ten months later, the patient was re-admitted to our department. A mucin-producing cauliflower-like mass was found in the epigastric region (Figure 1). The patient complained that the previous PCD wound on the abdominal wall would not heal, and jelly-like material was found there at first, after which a cauliflower-like mass grew from the tract wound, gradually enlarging. Biochemical tests were unremarkable, and alpha-fetoprotein (AFP), CEA and CA19-9 were within normal limits. Abdominal CT demonstrated a low-density irregular mass in the left lobe with enhanced incomplete septa, with part of the mass extending outside the abdominal wall (Figure 2).

From these findings, we concluded that the mass in the epigastric region was formed as the tumor metastasized through the PCD tract. At laparotomy, a large mass connecting with the cauliflower-like mass outside the abdomen was found in the left hepatic lobe; no infiltration was found around the mass. Complete resection of the tumor was achieved by a left hemihepatectomy with partial abdominal wall tissue.

The surgical specimen contained the left lobe with the cauliflower-like mass and partial abdominal wall tissue. There was a unilocular cyst measuring 6x8x5 cm^3 in the left lobe, and the internal cavity was filled with papillary tumors (Figure 3).

The microscopic image showed cystic structures of varying size containing mucinous fluid. The cystic structures were lined with atypical mucin-producing glandular epithelium; cells had a columnar shape and an increased nuclear/cytoplasmic ratio (Figure 4).

The patient had an uneventful postoperative course and remains asymptomatic two years postoperatively without disease recurrence.

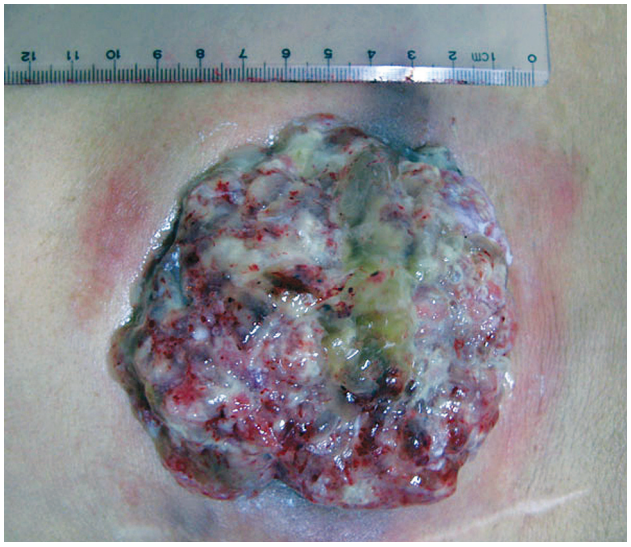

Figure 1. A mucin-producing cauliflower-like mass measuring 10x10x5 cm^3 in the epigastric region.

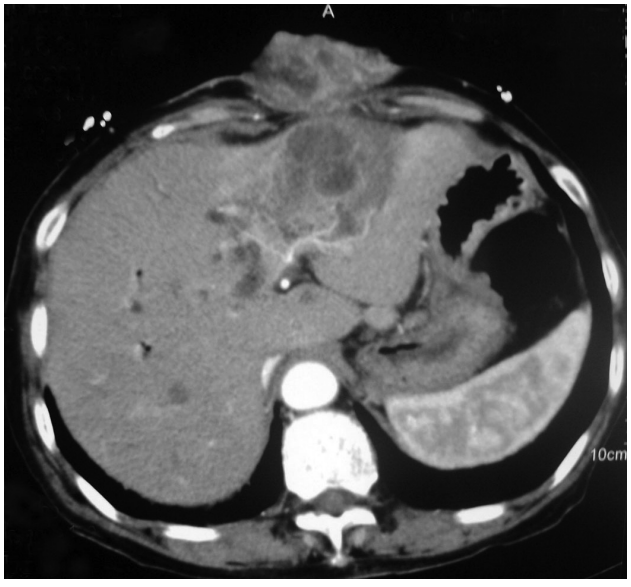

Figure 2. Enhanced computed tomography demonstrates a low-density irregular mass in the left lobe with enhanced incomplete septa, and part of the mass is seen to extend to outside the abdominal wall.

DISCUSSION

Biliary cystadenocarcinoma (BCAC) is a rare malignant epithelial tumor of the liver, with an incidence of 0.41% among all hepatic malignant epithelial tumors (3). It usually occurs in middle-aged women, and its etiology is unclear.

The exact diagnosis of BCAC is difficult during the preoperative course. The clinical presentation and radiological imaging are usually not distinctive

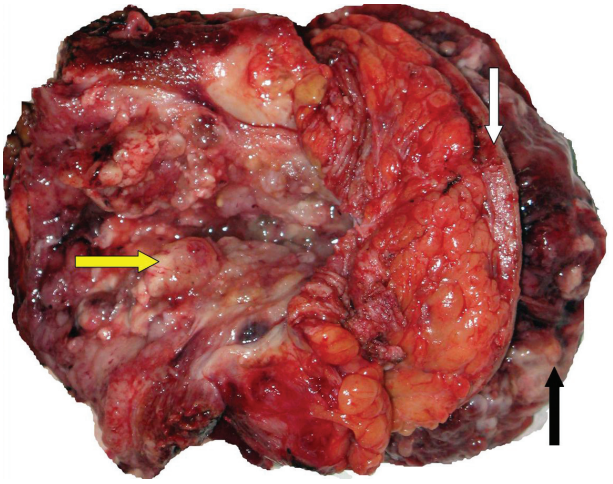

Figure 3. Gross appearance of the surgical specimen: left lobe with cauliflower-like mass (black arrow) and partial abdominal wall tissue (white arrow). A unilocular cyst measuring 6x8x5 cm^3 was seen in the left lobe, and the internal cavity was filled with papillary tumors (yellow arrow).

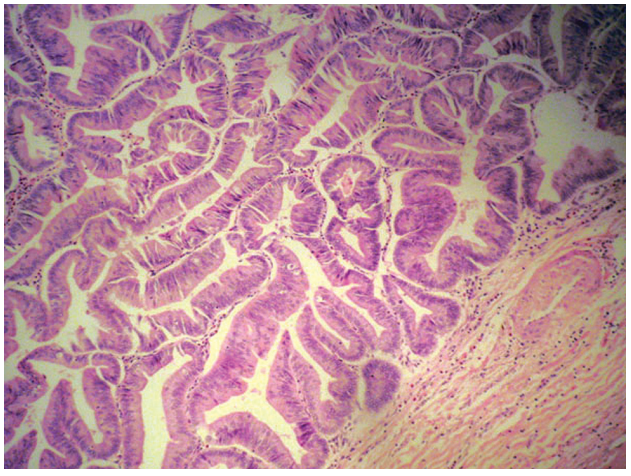

Figure 4. Microscopic image showing variously sized cystic structures containing mucinous fluid. Cystic structures were lined by atypical mucin-producing glandular epithelium; cells had a columnar shape and an increased nuclear/cytoplasmic ratio (H&E stain, x100).

from other lesions of the liver. The clinical presentation may include diffuse abdominal discomfort or severe constant pain in the abdominal region, a palpable abdominal mass, intermittent or constant jaundice, hepatomegaly, ascites, anorexia, vomiting, weight loss, and dyspepsia (4-6). The characteristic radiological features of BCAC are usually a solitary, large, well-defined multilocular cystic lesion. The thick fibrous capsule and internal septations are often visible and help distinguish BCAC from a simple cyst. Findings of internal septa with nodularity or mural nodule are indicative of BCAC. The presence of coarse calcifications along the wall and internal septum are also suggestive of BCAC (5,7,8), and after intravenous administration of contrast agent, enhancement of the septa and mural nodules can be seen in BCAC. However, about 20%-30% of liver abscesses have a septated or multilocular appearance on CT image, with or without calcifications (7,9). Hydatid cysts usually present with thicker tumor walls and, in some cases, with oval or round cysts (7,9). Rarely, cystic primary hepatocellular carcinoma or metastatic disease simulates a unilocular cystadenoma or cystadenocarcinoma (7,9). Therefore, BCAC is often misdiagnosed as a hepatic abscess, hydatid cyst, simple cyst, or metastatic tumor with cystic degeneration.

Patients with cystic liver disease are often treated by biopsy or by drainage of the cyst contents. Several cases of peritoneal carcinomatosis secondary to imprudent biopsy of the BCAC have been reported in the literature (10-12). PCD was performed in some patients with BCAC (13-15), either as drainage alone for relief of pain (15) or in combination with systemic chemotherapy and radiation (13) or infusion of 5-fluorouracil into the cyst (14). To date, no follow-up data were available from these patients. In this case, ultrasonography-guided PCD was performed on the basis of the initial incorrect diagnosis of liver abscess, and malignant seeding through the tract after this procedure was encountered. To our knowledge, this is the first reported case of a BCAC with malignant seeding through the PCD tract. The time interval between the procedure and diagnosis of needle tract implantation was 10 months, and the metastatic tumor measured 10 cm. Therefore, biopsy and drainage should be avoided in BCAC. Although the drainage may relieve the pain in poor surgical candidates (15), experience from this case has shown that rapid and aggressive growth of implanted tumors can occur in a short time period.

Total excision of a cystadenocarcinoma of the liver is the treatment of choice when feasible (5). Even though the metastatic tumor had reached a significant size in this case, the cystadenocarcinoma was still confined to the cystic lesions without invasion of peripheral organs in the abdominal cavity. Total surgical resection of the neoplasm with malignant seeding of the tract had a favorable outcome, and no local recurrence was found after a follow-up of more than two years, which implies its relatively benign clinical course.

In conclusion, PCD should be performed most ca-

refully in atypical liver cystic disease to prevent complications such as malignant seeding of the tract; however, if malignant seeding along the tract occurs in a patient with BCAC after PCD, complete excision of the primary and metastatic tumor may have good results.

REFERENCES

1. Yu Q, Chen T, Wan Y-L, et al. Intrahepatic biliary cystadenocarcinoma: clinical analysis of 4 cases. Hepatobiliary Pancreat Dis Int 2009; 8: 71-4.
2. Shrikhande S, Kleeff J, Adyanthaya K, et al. Management of hepatobiliary cystadenocarcinoma. Dig Surg 2003; 20: 60-3.
3. Takayasu K, Muramatsu Y, Moriyama N, et al. Imaging diagnosis of bile duct cystadenocarcinoma. Cancer 1998; 61: 941-6.
4. 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.
5. Läuffer JM, Baer HU, Maurer CA, et al. Biliary cystadenocarcinoma of the liver: the need for complete resection. Eur J Cancer 1998; 34: 1845-51.
6. Thomas KT, Welch D, Trueblood A, et al. Effective treatment of biliary cystadenoma. Ann Surg 2005; 241: 769-73.
7. Buetow PC, Buck JL, Pantongrag-Brown L, et al. Biliary cystadenoma and cystadenocarcinoma: clinical-imaging pathologic correlations with emphasis on the importance of ovarian stroma. Radiology 1995; 196: 805-10.
8. Pojchamarnwiputh S, Na Chiangmai W, Chotirosniramit A, Lertprasertsuke N. Computed tomography of biliary cystadenoma and biliary cystadenocarcinoma. Singapore Med J 2008; 49: 392-6.
9. Korobkin M, Stephens DH, Lee JK, et al. Biliary cystadenoma and cystadenocarcinoma: CT and sonographic finds. AJR 1989; 153: 507-11.
10. Nakajima T, Sugano I, Matsuzaki O, et al. Biliary cystadenocarcinoma of the liver: a clinicopathologic and histochemical evaluation of nine cases. Cancer 1992; 69: 2426-32.
11. Iemoto Y, Kondo Y, Fukamachi S. Biliary cystadenocarcinoma with peritoneal carcinomatosis. Cancer 1981; 48: 1664-7.
12. Wolf HK, Garcia JA, Bossen EH. Oncocytic differentiation in intrahepatic biliary cystadenocarcinoma. Modern Pathol 1992; 5: 665-8.
13. Kasai Y, Sasaki E, Tamaki A, et al. Carcinoma arising in the cyst of the liver: report of three cases. Jpn J Surg 1977; 7: 65-72.
14. Kanamori H, Kawahara H, Oh S, et al. A case of biliary cystadenocarcinoma with recurrent jaundice: diagnostic evaluation of computed tomography. Cancer 1985; 55: 2722-4.
15. Mauro MA, Parker LA. Percutaneous drainage of a cystic tumor for relief of pain. Southern Med J 1987; 80: 1466.