Doi: 10.32677/IJCR.2016.v02.i02.007 Case Report

# Biliary Cystadenoma - A Case Report

**Kopperundevi Vadamalai**

*From Department of General Surgery, Thanjavur Medical College, Thanjavur, Tamil Nadu, India*

**Correspondence to:** Dr Vadamalai Kopperundevi, T1, Vadivu Abodes, 29, Rahuman Nagar Main Road, Rahuman Nagar, Thanjavur, Tamil Nadu - 613004, India. E mail: drvkdevi@gmail.com.

*Received: 09 March 2016 Initial Review: 04 April 2016 Accepted: 14 April 2016 Published online: 23 April 2016*

## ABSTRACT

Biliary cystadenoma are rare cystic lesions of liver. They accounts for less than 5% of nonparasitic cyst of liver. Hueter first reported biliary cystadenoma in 1887. It is an extremely rare benign hepatic tumor arising from von meyerberg complexes. It occurs frequently in middle age between 40 and 50 years with female preponderance. We, hereby, report a case of 70 year male, who presented with mass and pain in the right upper abdomen.

**Keyword:** Liver, Cyst, Biliary cystadenoma, Biliary cystadenocarcinoma

Biliary cystadenoma is an extremely rare and benign hepatic tumor accounts for less than 5% of nonparasitic cyst of liver [1] with potential for malignant transformation [2]. It is difficult to distinguish biliary cystadenoma and biliary cystadenocarcinoma preoperatively and hence, surgical resection should be considered. Common location is intrahepatic (85%); out of which, 29% involve left lobe, 50% right lobe and 16% involve both the lobes. It usually occurs in females (female to male ratio - 4:1) and the age group between 40 and 50 years is commonly affected [3]. Origin of tumour is due to proliferation of ectopic embryonic tissues that otherwise aid in the development of adult gall bladder [4]. It usually presents late because of its slow growing nature.

## CASE REPORT

A 70 year old male presented with the mass in upper abdomen for past one month associated with vague abdominal pain, more on right side and early satiety. There was no history of jaundice. He had history of bull gore injury 30 years back for which laparotomy was done; however, the treatment records were not available.

No other relevant history was given by the patient. On general examination, he was moderately built and nourished, not icteric, and there was no pallor or lymphadenopathy seen. His vitals were stable. A mass of size 20x15cm, firm in consistency, was felt in right hypochondrium extending up to right iliac fossa (figure 1). The other systemic examinations were normal. Clinical differential diagnosis of hydatid hepatic cyst was made.

His routine blood investigations including complete hemogram, peripheral smear, and renal and liver function tests were within normal limits. Ultrasound examination of abdomen showed a cystic lesion in the right lobe of liver extending up to pelvis which was suggestive of biliary cystadenoma. CT abdomen also showed a cystic lesion in the right lobe of liver 20x10 cm size extending up to pelvis, with left diaphragmatic hernia (contents probably colon) confirming the diagnosis of biliary cystadenoma along with left diaphragmatic hernia (Figure 2). Upper gastrointestinal endoscopy revealed extraneous compression of the stomach.

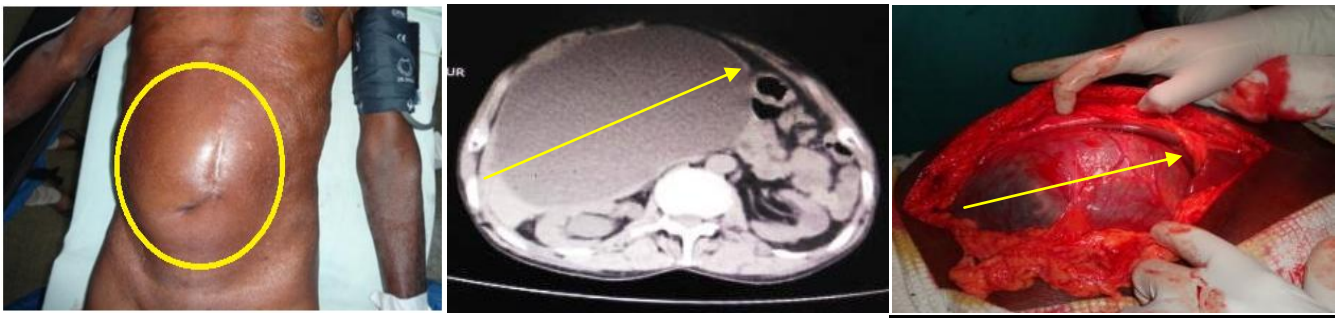

**Figure 1 - Preoperative picture showing mass in the abdomen occupying the right hypochondrium, epigastric, umblicus and right iliac fossa region. Figure 2 - CT Abdomen shows cystic lesion in right lobe of liver 200x100mm size extending up to pelvis. No septations seen. Figure 3 – Intra-operative picture showing the cyst before aspiration.**

The patient underwent laparotomy that revealed cyst of 20x10 cm arising from right lobe of liver. Complete resection of cyst was done after aspirating about 1.5 litres of clear fluid (Figure 3 and 4). Left diaphragmatic hernia repair was also done with mesh after reducing the contents (splenic flexure of transverse colon and spleen). Intra-operative and post-operative period was uneventful and intercostal drain was removed on 5$^{th}$ post operative day.

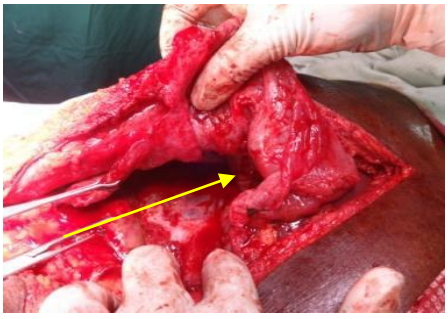

**Figure 4 – Intra-operative picture showing cyst wall after aspiration.**

Specimen was sent for histopathlogical examination which showed a cyst wall contains epithelial lining of biliary type non-ciliated columnar epithelium. There was no evidence of ovarian stoma or malignant changes. Patient continues to do well for the past 1 year with no signs of recurrence.

## DISCUSSION

Hepatobiliary cystadenoma can occur at any age but they are usually seen in middle aged women [3]. These tumours present commonly as multiloculated cyst while unilocular cystadenomas are rare [5]. Clinical presentation is usually with abdominal pain or discomfort, with abdominal distension and palpable mass [1, 3, 6]. Acute pain may occur due to rupture of the cyst or due to intracystic haemorrhage while infection of the cyst can cause fever [7]. Jaundice and ascites due compression of bile duct and inferior vena cava by the huge size of the cyst have also been reported [8,9,10]. On literature search, we could not find any report on cystadenoma causing diaphragmatic hernia. The cause of diaphragmatic hernia in our patient could be the previous injury. Differential diagnosis include parasitic cyst, liver abscess, cystic cholangiocarcinoma, and hepatobiliary mesenchymal tumours like biliary leiomyoma [3,11].

On ultrasound examination, cystic nature of the lesion, septations, internal echoes and papillary projection if present can be demonstrated [12, 13]. CECT demonstrates the anatomic relation to the surrounding structures, particularly major vessels. MRI defines the nature of fluid e.g. blood or mucin [14]. Multilocular cystic lesion with calcification along the septa indicates likely diagnosis of cystadenocarcinoma [15]. The presence of irregular thickness, mural nodule or papillary projection indicates the possibility of malignancy. Investigations like intra-operative cholangiography, and ERCP are indicated only if communication of the cyst is suspected.

Initial definition of cystadenoma by Edmondson in 1958 and by Wheeler & Edmondson in 1985, includes three distinctive features. The lesion should be i) Multilocular ii) Lined by a columnar epithelium iii) Accompanied by a dense cellular ovarian like stroma. Presently, there are divided in two types i.e. 1)

Hepatobiliary cystadenoma with mesenchymal stroma (ovarian like) and 2) Hepatobiliary cystadenoma without mesenchymal stroma. Hepatobiliary cystadenoma with ovarian stroma were exclusively seen in females and these are considered pre-malignant with good prognosis while cystadenoma without stroma are known to transform into malignancy more often with poor prognosis [6,12]. Cystic fluid may be clear, mucinous and rarely bile stained or purulent. Presence of blood stained fluid indicates malignant component [12].

Cystic fluid analysis for CEA, CA19-9 can be useful in differentiating cystadenoma from other hepatic cystic lesion. Elevated markers give indication of biliary origin of the cyst [1,7,12]. Only histopathological examination of the resected specimen is the final diagnostic method of cystadenoma and cystadenocarcinoma. If a cystadenoma is suspected, or has been diagnosed, surgery is indicated even in asymptomatic patients since cystadenoma and cystadenocarcinoma cannot be reliably differentiated based on the radiologic and macroscopic criteria. Partial excision and drainage may lead to recurrences [1,16,17].

## CONCLUSION

Biliary cystadenoma is a rare benign hepatobiliary tumour which mostly affects middle aged females and manifest as multi locular cystic mass. This case occurred in old age male with unilocular cystic lesion without mesenchymal stroma.

## REFERENCES


1. Wheeler DA, Edmondson HA. Cystadenoma with mesenchymal stroma (CMS) in the liver and bile ducts. A clinicopathologic study of 17 cases 4 with malignant change. Cancer. 1985; 56(6): 1434-45.
2. Ishak KG, Willis GW, Cummins SD, Bullock AA. Biliary cystadenoma and cystadenocarcinoma: report of 14 cases and review of the literature. Cancer. 1977; 39 (1): 322–38.
3. Florman SS, Slakey DP. Giant biliary cystadenoma: case report and literature review. American Surgeon. 2001; 67(8): 727–732.
4. Subramonyc, Herra GA, Turbatt-Herrera EA. Hepatobiliary cystadenoma: a study of five cases with reference to histogenesis. Arch Pathol Lab Med. 1993; 117: 1036-42.
5. Choi BI, Lim JH, Han MC, Lee DH, Kim SH, Kim YI, et al. Biliary cystadenoma and cystadenocarcinoma: CT and sonographic findings. Radiology. 1989; 171: 57-61.
6. 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(11): 1078–1091.
7. Dixon E, Sutherland FR, Mitchell P, McKinnon G, Nayak V. Cystadenoma of the Liver: a spectrum of disease. Canadian J Surg. 2001; 44(5): 371-376.
8. Preetha M, Chung AYF, Lim-Tan SK, Lim DTH, Thng CH. Intrahepatic biliary cystadenoma presenting with obstructive jaundice. Asian J Surg. 2004; 27(3): 243–245.
9. Catinis GE, Frey DJ, Skinner JW, Balart LA. Hepatic cystadenoma: an unusual presentation. Am J Gastroenterol. 1998; 93(5): 827–829.
10. Zhou JP, Dong M, Zhang Y, Kong FM, Guo KJ, Tian YL. Giant mucinous biliary cystadenoma: a case report. Hepatobiliary and Pancreatic Diseases International. 2007: 6(1): 101–103.
11. Palacios E, Shannon M, Solomon C, Guzman M. Biliary cystadenoma: ultrasound, CT, and MRI. Gastrointestinal Radiology. 1990; 15(4): 313–316.
12. Manouras A, Markogiannakis H, Lagoudianakis E, Katergiannakis V. Biliary cystadenoma with mesenchymal stroma: report of a case and review of the literature. World J Gastroenterol. 2006; 12(37): 6062–69.
13. Forrest ME, Cho KJ, Shields JJ, Wicks JD, Silver TM, Mc-Cormick TL. Biliary cystadenomas: sonographic-angiographic-pathologic correlations. Am J Roentgenol. 1980; 135(4): 723–727.
14. Federle MP, Filly RA, Moss AA. Cystic hepatic neoplasms: complementary roles of CT and sonography. Am J Roentgenol. 1981; 136(2): 345–348.
15. Koroglu M, Akhan O, Akpinar E, Oto A, Gumus B. Biliarycystadenoma and cystadenocarcinoma: two rare cystic liver lesions. JBR-BTR. 2006:89261–63.
16. Fiamingo P, Veroux M, Cillo U, Basso S, Buffone A, D'Amico DF. Incidental cystadenoma after laparoscopic treatment of hepatic cysts: which strategy? Surg Laparosc Endosc Percutan Tech. 2004; 14: 282-284.
17. Hai S, Hirohashi K, Uenishi T, Yamamoto T, Shuto T, Tanaka H, et al. Surgical management of cystic hepatic neoplasms. J Gastroenterol. 2003; 38(8): 759–764.


**How to cite this article:** Vadamalai K. Biliary Cystadenoma - A case report. Indian J Case Reports. 2016; 2(2): 36-38.

*Conflict of interest: None stated, Funding: Nil*