INTERNATIONAL JOURNAL OF MEDICAL AND APPLIED SCIENCES
E-ISSN:2320-3137

Case Report

BILIARY CYSTADENOMA MASQUADERING AS HYDATID CYST.

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Abstract :

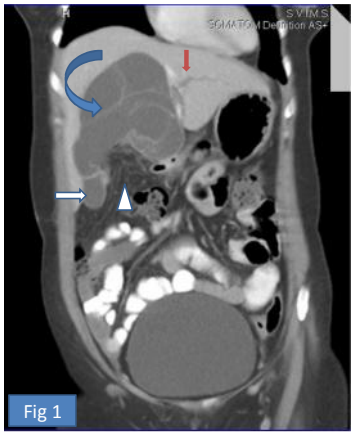
Biliary cystadenomas are rare cystic neoplasms of hepatobiliary epithelial origin usually intrahepatic, occurring predominantly in middle aged women. These usually present as large multi-loculated septated cystic lesion in liver. Hydatid cyst poses a great mimicker to biliary cystadenoma, sometimes making it impossible to differentiate on imaging alone. There have been fewer than 150 & 50 case reports of biliary cystadenoma & cystadenocarcinoma respectively in literature so far. Due to its rarity, features of biliary cystadenoma have been sparsely investigated. We report a case which was radiologically diagnosed as hydatid cyst, given albendazole for two weeks, underwent laparoscopic deroofing of cyst whose histopathology of the cyst wall suggested biliary cystadenoma. The patient was again subjected to computed tomography (CT) scan, taken up for hepatectomy with histopathology proving it to be a biliary cystadenoma. Any therapy short of complete excision leads to local recurrence and has a potential risk of malignant transformation. Due to similar imaging appearances between them, biliary cystadenoma should always be a close differential diagnosis to hydatid, especially in countries with a high incidence of hydatid disease.

KEYWORDS: Biliary cystadenoma. Hydatid cyst. Cystic. Liver. Lesion. Benign.

CASE REPORT:

A 28 years old woman presented to surgical clinic with complaints of continuous dull aching pain in right upper abdomen since 6 days. On examination, hepatomegaly present, approximately 10 cm below costal margin in mid clavicular line, smooth surface, firm in consistency, non tender. All the routine lab investigations including liver function tests were within normal limits. CT abdomen done elsewhere showed a complex cystic lesion with septations in left lobe , likely to be hydatid cyst. Patient was started on tab. Albendazole 400mg twice daily for two weeks and planned for laparoscopic deroofing of cyst. Intra-operative findings showed a multilocular cyst in segments IV and V of liver. Clear fluid with bilious tinge was present intra-cystically . Deroofed cyst wall was sent for histopathological examination which suggested mucinous cystic neoplasm / biliary cystadenoma. CT abdomen was repeated which showed a well defined hypodense lesion with enhancing peripheral wall and septae in segments IVa and IVb of left lobe extending into segment V and upto porta hepatis. Mild adjacent intra-hepatic biliary dilatation is noted in the left lobe (**fig.1**) with features suggesting biliary cystadenoma. Patient was readmitted and planned for left hepatectomy with resection of segment V. Intra operative findings showed a single cyst measuring 10 X 8 cms in segments IVa and IVb extending into segment V (**fig. 2**). Cyst

contents were serous with bilious tinge. Histopathology of resected specimen was reported as biliary cystadenoma.



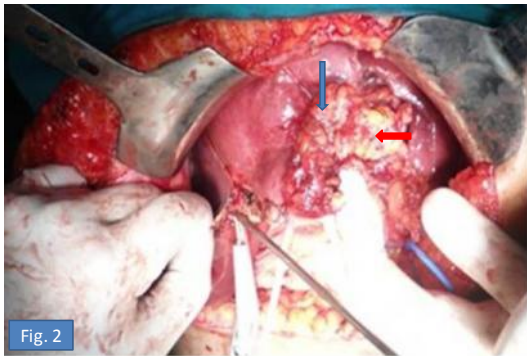


Fig. 1 : CECT coronal section of biliary cystadenoma showing multiloculated cystic lesion with enhancing wall and internal septae (curved arrow) in segments IV a and IV b, dilatation of intrahepatic ducts in left lobe (red arrow). Gallbladder displaced inferiorly (white arrow). Post operative changes following deroofing of the cyst (arrowhead).

Fig 2 : Intraoperative photograph showing lesion in segment IV (blue arrow), omentum attached to the lesion following previous deroofing of cyst wall (red arrow).

DISCUSSION :

Biliary cystadenomas are rare cystic neoplasms of hepatobiliary origin which constitutes less than 5% of intra hepatic cysts [1]. These usually present as large multi-loculated septated cystic lesions in liver, primary location being hepatic hilum in segment IV [2]. They have a high tendency to recur after palliative procedures and have potential risk for malignant transformation, making preoperative diagnosis imperative. Since, its imaging features are similar to hydatid cyst, preoperative differentiation poses a diagnostic challenge especially in countries with a high incidence of hydatid disease, sometimes impossible [3]. Due to its rarity, features of biliary cystadenoma have been sparsely investigated.

It is more common in middle aged females who present asymptomatically or with abdominal pain and or a palpable mass [4]. Our case is a 28 year old female from a hydatid endemic region of South India.

It is a true proliferative epithelial tumour with two histologic variants, a serous type and a far more commoner mucinous type. Cystadenoma with mesenchymal stroma is regarded as a precancerous lesion which occurs exclusively in women [4].

Most commonly used diagnostic tools to image biliary cystadenoma are ultrasonography and CT on which they appear as well defined ovoid hypoechoic/low density lobulated multi-loculated septated intrahepatic cystic lesions. Enhancement of the internal septae and walls are noted on contrast administration [5]. Upstream dilatation of adjacent intrahepatic biliary radicles can be seen due to mass effect . Demonstrable communication of the cyst with biliary system is specific, but is rarely seen. Differential diagnosis of cystic hepatic lesions

include liver cysts, hydatid cysts, hematomas, mesenchymal hamartomas, liver abscess, congenital cysts, polycystic disease, caroli's disease, neoplastic lesions such as biliary cystadenocarcinoma, undifferentiated embryonal sarcoma and cystic metastasis. Among these, liver abscess and hydatid disease are the two entities most likely to be confused with biliary cystadenoma [6]. Thick irregular walls with internal echoes are features of abscesses while crenated margins favour amoebic etiology. Hydatid liver cysts caused by echinococcus granulosus are commonly multilocular because of the presence of daughter cysts within the original cyst. Usually, type CE 2 cyst by WHO 2001 classification appearing as multivesicular cysts arranged in the form of rosettes pose a diagnostic challenge in differentiating them from biliary cystadenoma. [7]

Total resection of the lesion with a prolonged close follow-up is the management of choice of biliary cystadenoma due to high possibility of local recurrence after palliative procedures and potential risk of malignant transformation into cystadenocarcinoma [4].

Hence, preoperative differentiation from other cystic lesions is substantially important. Imaging features favouring cystadenomas over hydatid are multiloculated, usually single, cystic intrahepatic lesions in middle aged women, in left lobe, with upstream dilatation and demonstrable communication of the cyst with adjacent intrahepatic biliary radicles. Due to similar imaging appearances, biliary cystadenoma should always be a close differential diagnosis to hydatid, especially in countries with a high incidence of hydatid disease [3].

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