

Biliary Cystadenoma -A Diagnostic dilemma: A Case report

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Abstract

Biliary cystadenomas is an uncommon benign cystic neoplasm of the liver and occur predominantly in middle-aged patients and are more common in women¹. There is a difficulty in differentiating a benign from a malignant biliary cystadenoma and hence many methods like Ultrasound, CT or MRI are often used in its diagnosis. Biliary cystadenomas often cause a diagnostic dilemma with other cystic lesions, especially hydatid cysts in endemic regions. In the present case report, a case of a biliary cystadenoma in a young female patient who presented with an upper abdominal swelling is reported.

Key Words : Biliary cystadenoma, Liver cyst

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Introduction

Biliary cystadenomas constitute less than 5 % of all hepatic solitary non parasitic cysts [1]. It is a benign tumor, although prone to malignant degeneration [2], supposedly originating in intrahepatic or rarely extrahepatic embryonic tissue precursors of biliary epithelium [3]. Hueter first reported BCA in 1887 & Keen reported the first BCA resection 5 years later [4, 5]. Because of the rarity, many clinicians are unfamiliar with the diagnostic features and therapeutic management. It is often misdiagnosed as a hydatid cyst, especially in places where parasitic cysts are very common. We present the case of a biliary cystadenoma in a 22 year old female patient who presented with an upper abdominal swelling.

Case report

A 22 year old female patient presented with upper abdominal pain and distension. She had no history of oral contraceptive intake. On examination, there was a large swelling (18 x 15)cm in the right hypochondrium, cystic in feel, slightly tender and moving with respiration. Liver function tests and full blood counts were normal. Anti - echinococcal antibodies were negative. Ultrasonography of the abdomen showed a well defined cystic lesion arising from the right lobe of the liver and about 15 cm in maximum diameter. CT scan could not be done due to financial constraints. A provisional diagnosis of hydatid cyst was made. The patient underwent midline laparotomy. There was a large cystic lesion arising from the inferior surface of the right lobe of the liver. It

contained clear mucinous fluid. The cyst was completely excised. On opening the cyst, multiple small cystic projections (Figure-1) were seen lining the inner wall. The cyst wall was sent for biopsy. Histopathological examination showed a cystic space lined by tall columnar mucin secreting epithelium with basal nuclei. Cyst wall showed hyalinization. It was diagnosed to be a case of biliary cystadenoma of the mucinous variety. Post operative period was uneventful and the patient is currently under follow up.



Figure-1: Photography depicting Biliary Cystadenoma

Discussion

A biliary cystadenoma is a cystic lesion which constitutes less than 5 % of all hepatic solitary non parasitic cysts. Almost 95% occur in women, the mean age being 45 years (range 2 – 87 years) [6]. The almost exclusive female predominance suggests a strong hormonal influence [7]. Although it occurs mainly in middle age, our patient was a young female. Biliary cystadenomas are usually located in the right lobe of the liver(50%), although they may also be present in both lobes (16%) or only in the left lobe of the liver (29%). Though some unilocular cystadenomas have been reported, multilocular ones are more

common [3]. Patients usually present with a slow growing abdominal swelling. Other symptoms are pain, anorexia, nausea, weight loss and ascitis [1,3]. Other less common symptoms include painful intracystic haemorrhage, rupture and fever from secondary infection [8]. Jaundice by compression, protrusion, invasion of bile ducts or by secretion of dense mucinous material has been reported [9]. Invasion of the bile ducts may result in episodes of pancreatitis [3]. Cystadenomas are known to increase in size during pregnancy and following use of oral Contraceptives, suggesting hormonal dependence [10-12].

The cyst fluid is usually clear and mucinous. Blood stained fluid indicates a malignant component [13]. Differential diagnosis of cystadenomas include simple liver cysts, parasitic cysts (particularly hydatid cyst), haematomas, post traumatic cysts, liver abscesses, polycystic diseases, embryonal sarcoma, primary or metastatic necrotic neoplasm, biliary intraductal papillary mucinous neoplasm and biliary cystadenocarcinomas [13-16]. Diagnosis and differential diagnosis of cystadenomas from other hepatic cystic liver lesions is mainly based on abdominal US, CT and MRI. Ultrasonography has proven very useful as an initial investigation as it outlines the anechoic mass with thin internal septa that are highly echogenic. Some internal echoes may represent papillary growth instead of septation [1,8]. CT & US are complementary modalities in evaluating BCTs. Sonography is more sensitive for detecting septa in cystic lesions & CT more

accurately demonstrates the site and anatomic extent of these lesions. CECT in addition demarcates the anatomic relation to surrounding structures, particularly major vessels [13,16]. MRI is able to characterise the nature of fluid within the cyst , that is, blood versus mucin (13, 14). The presence of irregular thickness of the walls, mural nodules or papillary projections indicates the possibility of malignancy [1,14,15]. Core needle biopsy for diagnosis risks dissemination of tumor cells and is not recommended (1, 8).

Biliary cystadenomas have historically been treated by marsupialization, internal Roux drainage, aspiration, sclerosis or partial resection [17]. All of these methods have been associated with high rates of complications including sepsis, rupture, haemorrhage, continued growth and progress to malignancy [1]. Fenestration and complete fulguration of the cystic bed has also been reported in the literature as a successful treatment option but this remains yet to be confirmed whether it is indeed successful [17]. Whenever a cystadenoma is suspected or diagnosed, surgery is indicated even if the patient is asymptomatic. Ideal treatment is radical excision [1]. In case of bilioenteric communications, closure of the fistula should be ensured.

To conclude, the diagnosis of biliary cystadenoma should always be kept in mind when we encounter multilocular cystic lesions of the liver in females, especially in areas where hydatid cyst is endemic. Complete excision

should always be the treatment of choice.

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Biliary Cystadenoma -A Diagnostic dilemma: A Case report

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