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Case Report

Hepatic Hydatid Diagnosed Before Operation but Mucinous Biliary Cystadenoma of Intrahepatic Bile Duct By Pathology after Operation: A Case Report

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1. Abstract

Intrahepatic biliary cystadenoma is a rare benign tumor of the liver, with less than 5% incidence. Although with the progress of imaging, the discovery of this disease species is more than before, but because this disease has no specific diagnostic means, the preoperative misdiagnosis rate is very high. We report a case of hepatic hydatid diagnosed before operation but mucinous biliary cystadenoma of intrahepatic bile duct by pathology after operation.

2. Introduction

Intrahepatic biliary cystadenoma is a rare benign tumor of the liver, with less than 5% incidence. Although with the progress of imaging, the discovery of this disease species is more than before, but because this disease has no specific diagnostic means, the preoperative misdiagnosis rate is very high. We report a case of hepatic hydatid diagnosed before operation but mucinous biliary cystadenoma of intrahepatic bile duct by pathology after operation.

3. Case Report

The patient, a 32-year-old female, was admitted to a local hospital due to recurrent upper abdominal discomfort combined with post-meal nausea and vomiting. Abdominal ultrasound was performed at the local hospital, indicating hepatic echinococcosis, and the patient was referred to our hospital. The patient was healthy, had no smoking and drinking history, and had a pastoral life history. Enhancement CT (Figure 1) of our hospital diagnosed as large cystic lesion with multiple ascus, liver echinococcosis not excepted. Laboratory examination showed no significant abnormalities in liver function and blood routine, and CA199 was 65 mmol/L.

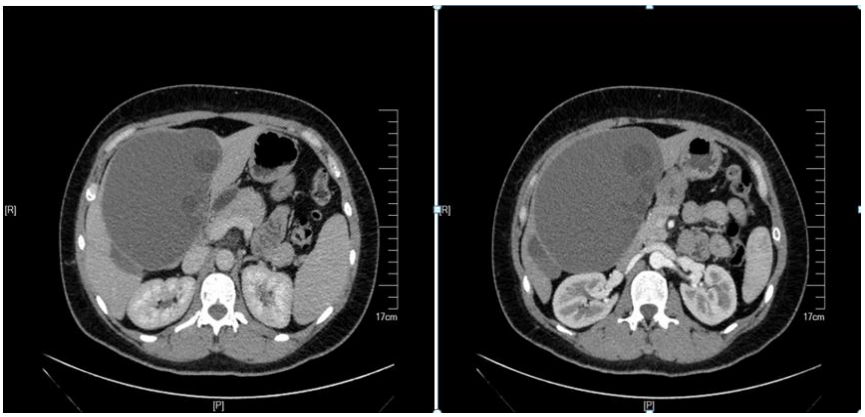

Figure 1: Enhancement CT found a nearly-circular cyst with low density at the left lobe of liver, of which size was about 15.1 x10.1 x14.3 cm and CT value was 14 to 22 hu. Multiple pockets lesions of low density can be seen in the huge lesion, part of which showed calcification. And enhancement scanning showed mild strengthening of the cyst wall.

3.1. Intraoperative Findings

All tumors' sites were consistent with preoperative ultrasound and CT localization. The lesion was compressed the gallbladder to the right , compressed the stomach to the left, and also the head and body of the pancreas was compressed, extended to the renal hilum. Lesions was adhered to visceral surface of liver .The base is located in the liver III, IV, V, V section of visceral face, close to the first porta hepatis. Macroscopic observation found the neoplasm wall was translucent, inflammatory thickening which is not like hydatid. Intraoperation into the mass cavity (Figure 2): there was lots of ball-like organization of different size in it. We excision the mass and part of liver along the mass envelope. Pathological diagnosis (Figure 3) is mucinous biliary cystadenoma.

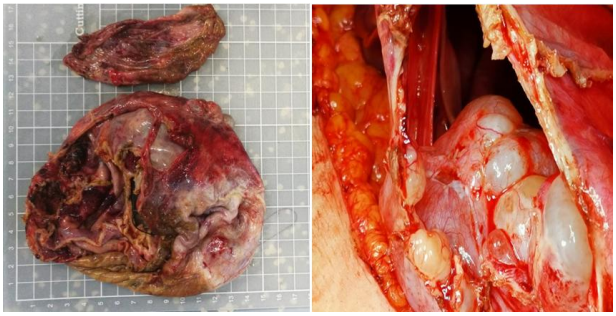

Figure 2: Postoperative pathological findings were as follows :(liver) one cystic mass, volume 8x6x4cm, the thickness of wall was 0.3cm, inner wall was light brown and brown; local bubblelike protrusion was visible, volume 4.5x4.5cmx3cm, the thickness of wall was 0.5cm , inner wall was light brown;Three vesicles could be found locally, whose diameter was about 0.5cm-1.2cm, contenting clear liquid.

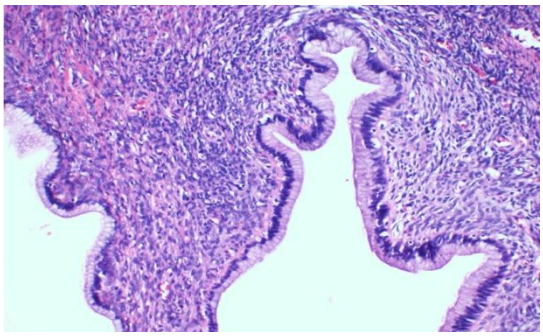

Figure 3: The H&E staining of the pathological section at *400:The walls of the cysts are covered with numerous monolayer mucinous columnar epithelium.

4. Discussion

Intrahepatic biliary cystadenoma is a kind of rare benign tumor of hepatic gland epithelium, the incidence of which is less than 5% [1, 2]. Most of them occur in the liver, while those occurring in the extrahepatic biliary duct are rare [3]. Some articles pointed out that the disease has a significant gender tendency, about 90% of the patients are female, and the average age of onset is about 45 years old [4, 5].

The patient often consults a doctor with abdominal mass, abdominal pain, other rare symptoms were dyspepsia, loss of appetite, disgusting, vomit, also can cause jaundice because of tumor's oppression or growing into the bile duct [6, 7]. Imaging examination often showed hepatic duct cystadenoma as isolation mass in liver, can also be multiple; It can be as single room, or a room with cyst or solid. It has complete capsule, but was unable to determine the nature of a cystic lesion, neither to identify with other liver cystic, which may lead to misdiagnosis, or under diagnosis, so surgery should be perform to definite pathological diagnosis. Because cystadenomas are often misdiagnosed as common cystic lesions, they are often treated with simple drainage or incomplete resection, which often results in recurrence or cystic swelling [8]. In this case, the imaging of the patient indicated slight enhancement of the internal cystic wall of the lesion, and multiple small sac-like low-density shadows within the lesion, presenting the appearance of sacs-in-sac. Combined with the patient's life history in the pastoral area, the initial diagnosis was hepatic echinococcosis. However, the intraoperative findings were significantly different from the imaging findings, indicating that the imaging findings of this tumor may be atypical. Thankfully, because of the rarity of the disease, biliary cystadenoma is being studied from a genetic, surgical, and radiological perspective [4].

BMCNs have been reported to increase in size during pregnancy and with oral contraceptives, suggesting hormonal dependency [9]. It has been proposed that KRAS gene mutation may be the main driving factor for biliary cystadenoma [10], which provides us with new ideas for the prevention and treatment of this tumor. After reading through literature review, there was only one previous case that liver hydatid disease was considered before surgery, but the postoperative pathological result was biliary cystadenoma [11]. This case has certain guiding significance in our clinical work.

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