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CASE REPORT

Ciliated Hepatic Foregut Cyst

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Ciliated hepatic foregut cyst is a rare, benign, most often solitary and unilocular, rarely multilocular cyst made up of a ciliated pseudostratified columnar epithelium, a subepitheial connective tissue layer, a smooth muscle layer and an outer fibrous capsule. The lesion is usually found incidentally by ultrasonography, during surgical exploration or autopsy. Recent publications characterizes of its fine needle aspiration biopsy features. The lesion is mostly asymptomatic, however one case caused portal vein compression and another which showed malignant trasformation through squamous metaplasia which warns to examine these lesions cautiously. As the lesion is extremely rare it is difficult to estimate its prevalence and its nature, so every single case presentation could be important. (Pathology Oncology Research Vol 8, No 4, 278–279)

Keywords: Liver, benign hepatic cysts, ciliated hepatic foregut cyst

Introduction

The ciliated hepatic foregut cyst (CHFC) is a rare, solitary mostly unilocular cystic lesion which derives from the embryologic foregut. The first descriptions of the lesion appeared from Friedrich[1] in 1857 however the term ciliated hepatic foregut cyst was first used by Wheeler and Edmondson[7] in 1984. Since 1988 numerous cases have been reported in Japanese patients. It is unclear whether these cases reflect an increased prevalance in Japanese patients compared with other populations or whether the Japanese medical communitiy has been more aggressive in reporting CHFC.[5] The cyst consists of four layers: ciliated pseudostratified columnar epithelium with admixed mucous cells, loose subepithelial connective tissue and a smooth mucle layer with an outer fibrous capsule. According to the comprehensive article of Vick and Goodman[5] the CHFC can be found in either of major lobes of the liver but occurs most often in the medial segment of the left lobe. The cysts typically appears in 50 years old patients with a slight male predominance. CHFC were originally found incidentally at autopsy, but later on the advent of ultrasound and CT imaging and the fine needle aspiration biopsy the lesion could be diagnosed precisely.

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Case report

Our single case is an incidental autopsy finding in a 55 years old male patient who died from hypertensive heart failure. The cyst was situated subcapsularly under the diaphragmatic surface of the right lobe near the insertion of the falciparum ligament of the liver. Its diameter was 2.5 cm, the wall thickness was a 1-2 mm. It was filled with a thick, yellowish, turbid mucoid material. The curiousity of the cyst content gave the idea of sampling the cyst for further examinations. The HE-stained sections showed an epithelial lining of ciliated pseudostratified columnar cells with occasional gobelt cells (*Figure 1–2*). Neither signs of

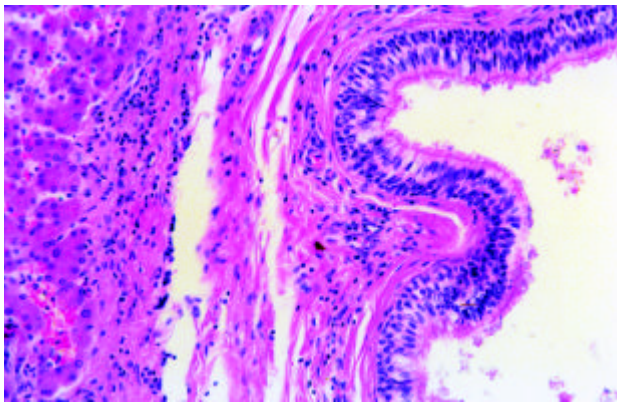

Figure 1. The epithelial lining, the smooth muscle wall and the surrounding liver tissue. HE, X100

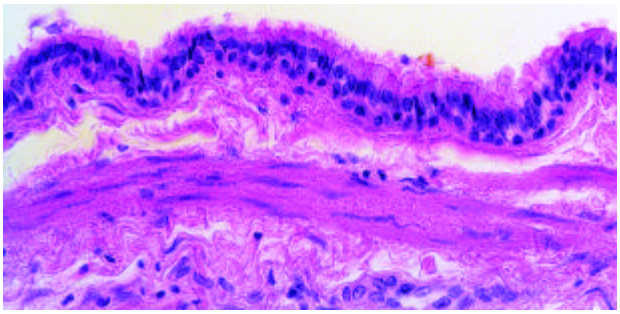

Figure 2. The ciliated epithelium and the smooth muscle wall on higher magnification. HE, X200

squamous metaplasia nor signs of malignancy could be observed in the epithelium. Underneath the epithelium a loose layer of conective tissue, a prominent layer of smooth muscle cells and an outer layer of connective tissue were seen. The goblet cells stained positively with periodic acid-Schiff and alcian blue.

The epithelial layer stained diffusely and strongly with cytokeratin and within the epithelium scattered Clara cells stained with chromogranin. The muscle fibers showed strong positive staining with smooth muscle actin.

Discussion

The ciliated hepatic foregut cyst is an extremely rare entity, which is mostly solitary, unilocular and bening in nature. The single malignant case warns to treat this entity cautiously.[6] The differential diagnostic possibilities includes the simple (cholangiogenic) cyst, parasitic (echinococcal) cyst, hepatobiliary cystadenoma and the cystic metastatic tumors. The radiology of these lesions can be critically important. As the lining epithelium is able to secrete a varity of fluids of variable viscosity, the density may therefore vary from near water to viscous or mucous. Calcification may also occur. The anatomic location of the cyst radiologically is probably the most important diagnostic consideration, as the cysts are always situated in a subcapsular location, on the anterior aspect of the liver, at the insertion of the falciparum ligament.[3,4] The fine needle aspiration biopsy appears to be completely diagnostic.[2,8] As the CHFC is the only known ciliated lesion in the liver, the presence of the tall, columnar epithelial cells with basally orientated nuclei and prominent apical terminal plates with cilia admixed with hepatocytes and mucous cells on aspiration biposy gives the diagnostic clue to this lesion.

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