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Malignancy of intraductal papillary neoplasm of the bile duct

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Dear Editor:

The intraductal papillary neoplasm of the bile duct (IPNB) is an uncommon disease which was first included in the World Health Organization classification of neoplasms in 2010. A 64-year-old woman was admitted to the hospital because of a hepatic lesion incidentally diagnosed during acute cholangitis. Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) (Fig. 1A) showed a well delimited 70mm mass, with a predominant cystic component and hyperenhancement of papillary internal branching, consistent with hydatid cyst. However, malignancy could not be excluded. The patient rapidly developed an acute abdomen syndrome, thus precluding liver biopsy. A new urgent CT was done to rule out a complication of the cystic lesion (Fig. 1B). A left hepatectomy was performed and the anatomopathological study confirmed the diagnosis of IPNB with a foci of cholangiocarcinoma therein (Fig. 1C and 1D). During follow up, the patient developed peritoneal carcinomatosis, received palliative chemotherapy and finally died.

Discussion



IPNB may be an incidental finding on image studies or could onset as a biliary obstruction. The most common imaging findings are intraductal masses with dilation of bile ducts. Despite the high risk of malignancy, this entity shows improved prognosis as compared with classical papillary cholangiocarcinoma. Differential diagnosis is challenging and may require pathological examination including the presence of mucin, papillary cells morphology and immunohistochemistry (1,2). Targeted biopsy specimens may be obtained by cholangioscopy (SpyGlass[®]) (3) and the only curative treatment is radical surgical resection.

In conclusion, IBPN may be suspected in patients with hepatic hilar lesions. A differential diagnosis is required with hydatid cyst, complicated cystadenoma or metastases, and it may prove difficult given the low specificity of imaging findings. A delay in the diagnostic process could allow for rapid clinical deterioration and death.

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Figure 1. A. Magnetic resonance cholangiopancreatography showing a 73x67 mm hilar hepatic space occupying lesion, hypovascular abd very heterogeneous in all MRI sequences. Areas with liquid signal, pseudonodular images and laminar pattern in T2 sequence. Diffusion restriction is seen in intermediate-intensity areas. B. Urgent intravenous contrast computed tomography. Cystic lesion growth, with increased density areas and some peripheral septa. Simultaneous proximal and distal bile duct dilation due to communication with the biliary duct. Ipsilateral portal vein thrombosis, liver microabscesses and free intraperitoneal fluid. C. Histological image showing intraductal papillary and pseudopapillary projections, with high-grade epithelial dysplasia. D. CK7+/20- tumour immunophenotype, consistent with biliary origin.