



Villous Adenoma of the Common Bile Duct Transforming into a Cholangiocarcinoma

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Villous adenomas of the upper gastrointestinal tract are rare [1], residing mostly in the duodenum (80%) and ampulla of Vater (15%). Villous adenomas of the extrahepatic bile ducts are exceptional and only a few cases have been reported in the medical literature worldwide [2]. The adenomatous polyp to carcinoma on the large bowel sequence is well known. However, in the absence of evidence of this sequence in the biliary duct a conclusion cannot be drawn from one case report on the adenoma-carcinoma sequence. We present a case of cholangiocarcinoma of the common bile duct arising from a villous adenoma.

Patient Description

A 77 year old woman with a history of recurrent abdominal pain and disturbed liver functions was admitted to the surgical

department because of obstructive jaundice and fever. One month previously an abdominal ultrasound examination showed an intracholedochal lesion with dilated intrahepatic bile ducts, dilated proximal common bile duct and enlarged lymph nodes in the hilum of the liver [Figure].

Physical examination revealed jaundice, tenderness in the right upper quadrant and mild hepatomegaly. Hemoglobin was 11.2 g/dl and white blood cell count $15 \times 10^9/L$; total bilirubin was 7.6 mg/dl and direct bilirubin 5.9 mg/dl. Liver enzymes were as follows: alanine aminotransferase 89 IU/ml (normal <40 IU/ml), aspartate aminotransferase 93 IU/ml (normal <40), alkaline phosphatase 791 IU/ml (normal <125), and gammaglutamyl transferase 744 IU/ml (normal <50). Endoscopic retrograde cholangiopancreatography was performed, revealing a dilated proximal CBD and

intrahepatic bile ducts. There was no evidence of sclerosing cholangitis or cystic disease of the biliary tract. A biopsy was taken from a suspicious stricture in the mid-portion of the CBD. The histologic examination showed a villous adenoma without dysplasia. There was one atypical gland in the stroma suspicious of malignancy. Following the finding of a dilated gallbladder on laparotomy, cholecystectomy and choledochotomy were performed. In the CBD at the level of the cystic duct insertion a villous tumor of approximately 2.5 cm in diameter was found. The CBD was resected 1 cm above and below the tumor and a Roux-en-Y hepaticojejunostomy was performed. The postoperative course was uneventful. The final histopathologic examination showed a tumor of 1.1 cm with

 CBD = common bile duct



[A] Axial ultrasound image revealing an hyperechoic intracholedochal lesion. [B] Ultrasound image obtained along the choledochus demonstrates a tubular hyperechoic lesion.

granular surface, leading to the diagnosis of a papillary well to moderately differentiated adenocarcinoma, probably arising from a villous adenoma [Figure B]. The surgical margins were free of tumor and the regional lymph nodes showed only atypia.

Comment

Benign biliary tumors are rare, comprising only 6% of all extrahepatic tumors. Two-thirds of these tumors are adenomatous papillomata or villous papillomata, and half of them are located at or close to the ampulla of Vater. Most cases are not diagnosed until surgery. They may be suggested preoperatively by ultrasound, ERCP and endoscopic ultrasound [2]; and suspected tissue specification can be obtained using an ultrasound-guided core needle biopsy or brushing via ERCP. The risk of malignancy is not well established, but it is generally considered that villous adenomas of the biliary tract probably behave like those found elsewhere in the gastrointestinal tract. For the colon and

rectum there is convincing evidence for malignant potential. Whether some carcinomas in the biliary tree arise from papilloma, as has been suggested, is not clear.

For benign lesions a complete excision with a cuff of normal bile duct is the treatment of choice [3]. Local excision by ERCP or laparotomy [2] has been proposed but the risk of recurrence is high. For malignant lesions the proposed treatment is hilar resection for proximal lesions and duodenopancreatectomy for distal ones [4]. Some authors advocate preoperative chemoradiation to achieve tumor-free resection margins using 5FU and 45 GY radiation of the porta hepatis [5]. A recent prospective study showed that postoperative radiotherapy does not improve survival in extrahepatic cholangiocarcinoma. In our case the tumor was located in the mid-part of the CBD; we performed resection of the CBD, including regional lymph nodes, with free margins, and Roux-en-Y hepaticojejunostomy. We believe that in the event of cholangiocarcinoma in the mid-part of the CBD, resection with free margins of the CBD with lymph node dissection of the

hepatoduodenal ligament is an appropriate treatment, but this should be confirmed by examination of large series.

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