Villous adenoma of the common hepatic duct: the role of ultrasound in management

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Abstract

A case of villous adenoma of the common hepatic duct causing obstructive jaundice, where the diagnosis was made by ultrasound guided percutaneous biopsy is reported. At surgery ultrasonography was used to define the extent and operability of the tumour.

Case report

A 58 year old man presented with a two month history of increasing painless obstructive jaundice. His alcohol consumption was 6 units per week. Investigations showed the serum bilirubin was 132 mol/l, the alkaline phosphatase 410 IU/l, and the alanine transaminase 36 IU/l. Tests for viral hepatitis were negative. Initial ultrasound scan (Acuson 128, 3.5 mhz) showed dilatation of the intrahepatic biliary tree and a 3 cm well defined, rounded mass at the hilum within the common duct. The mass showed no signs of infiltrating liver substance. A provisional diagnosis of benign tumour was made.

ERCP showed a normal pancreatic duct but the common bile duct could not be cannulated. A percutaneous transhepatic cholangiogram was therefore performed which confirmed a dilated intrahepatic biliary tree and a lobulated mass in the common hepatic duct (Fig 1). An 8 FG catheter was placed in the duct across the tumour to provide temporary biliary drainage. Subsequent ultrasound examination showed persistent dilatation of ducts within the right lobe of the liver although the left lobe was decompressed, implying involvement of the bifurcation of the common hepatic duct. Ultrasound guided 18 swg needle core biopsy of the tumour was then performed using the Biopsy TM system (Radimedical Ltd) described by Lindgren.¹ Histology of the core showed portions of a villous adenoma with mild epithelial atypia (Fig 2). There was no evidence of invasion. At surgery ultrasound examination (Hitachi UB30, 5 mhz intraoperative probe) showed a smooth tumour on a stalk lying free within the common hepatic duct and its principal branches with no signs of invasion (Fig 3). The common hepatic duct was opened, the tumour was enucleated and its stalk resected from the wall of the origin of the right hepatic duct.

Choledochoscopy was performed to ensure complete clearance of the ducts before closing. Histology of the surgical specimen confirmed the initial biopsy diagnosis with no evidence of stalk invasion (Fig 4). The patient made an uneventful recovery, remained well and was followed up with regular ultrasound scans. Sixteen months after the initial presentation a nodule of soft tissue density was noted on ultrasound at the bifurcation of the hepatic duct. An ERCP was

performed which showed a polypoid mass and biopsies confirmed recurrent villous adenoma. The lesion was again resected but on this occasion a generous length of bile duct was removed to reduce the chance of recurrence. A Roux en Y anastomosis was made to provide biliary drainage. The patient made a good recovery but will be monitored closely for recurrence.

Discussion

Villous adenomas are benign epithelial lesions with malignant potential which can occur at any site in the gastrointestinal tract. They are produced by a proliferation of the surface epithelium in the pattern of densely packed, thin, delicate fronds which are joined at their bases. They are usually encountered in the rectum and colon, less frequently in the small bowel and very rarely in the biliary tree.²

All types of benign neoplasm of the biliary tree are very uncommon. Only four cases were found by Burhans and Myers in a review of 4000 consecutive biliary tract operations.³ This brought the total of reported cases to 88 before 1972. Since then only a handful of cases have been reported in the literature. Most of the benign tumours described are derived from an epithelial or glandular origin and usually classified as papillomas, adenomas, cystadenomas, or

Figure 1: Percutaneous transhepatic cholangiogram showing lobulated mass obstructing common hepatic duct

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Figure 2: Intraoperative ultrasound scan showing clearly defined intraductal tumour.

simply polyps. The remainder consist of very small numbers of lipomas, myxomas, neuromas, and mixed tumours (adenomyomas, adenofibromas). The major duodenal papilla is the most frequently reported site of occurrence but many of these tumours may in fact have arisen from duodenal mucosa and are thus not true bile duct tumours. On review of the literature it becomes clear that there has been little uniformity in the nomenclature applied to the benign epithelial lesions and various classifications have been proposed^{4,5}. Hence lesions of apparently similar histological appearance have been given different labels by different workers. For example, villous adenoma is often recorded as papilloma or mucoid polyp and further confusion is caused by the differentiation of hyperplasia of normal papillary folds in the region of the ampulla from true epithelial tumours.^{4,6} In view of this, any hard data on frequency of occurrence of the types of benign tumour must be viewed with circumspection. Jaundice, pain and dyspepsia are the most common presenting features of benign biliary tumours.⁷ The tumours rarely grow large enough to become palpable because of their site.⁸ The degree of jaundice has been described to

Figure 3: Photomicrograph of core biopsy specimen showing papillary tumour with mild focal epithelial dysplasia but no evidence of malignancy. (H&E – medium power.)

Figure 4: Photomicrograph of operative specimen showing villous tumour with arborescent fibrous tissue core. No features of malignancy. (H&E – low power.)

fluctuate in some reports which may be attributable to a ball valve effect of the tumour. Most diagnoses are not made until surgery. The diagnosis of benign biliary tumour may be suggested pre-operatively by a combination of ultrasonography and cholangiography. The clearly defined nature of the lesion and absence of invasive features will differentiate it from malignant conditions such as cholangiocarcinoma. A cystic appearance with the presence of multiple septations may suggest a diagnosis of cystadenoma.⁸ Using the technique of ultrasound guided needle core biopsy with the Biopsy TM system a high degree of accuracy can be achieved with the added benefits of specific tissue characterisation. Potential complications of percutaneous core biopsy at this site include bile leakage and local haemorrhage although in a large recently reported series the actual major complication rate was low, at 0.5%.⁹

We believe this to be the first recorded case of biliary villous adenoma diagnosed by percutaneous biopsy. Despite only segmental decompression achieved by transhepatic drainage the benign appearance and biopsy results prompted surgical exploration with a view to curative resection. Most authorities would consider the treatment of choice for solitary benign tumours of the biliary tree to be local excision, with a tumour free margin to minimise the possibility of recurrence.^{10,11} In one series, however, operative mortality was similar for local and extensive excision but the recurrence rate was four times higher in the local excision group.³ Intraopera-

tive ultrasound is a well proven technique to aid accurate tumour resection and was used in this case to exclude an invasive component to the tumour. The risk of malignant change is not precisely known but it is generally considered that villous adenomata of the biliary tree behave like those elsewhere in the gastrointestinal tract. In the rectum and colon there is much convincing evidence that benign epithelial tumours have malignant potential.¹² The frequency with which invasive carcinoma is found increases with the size and number of the lesions.

There is general agreement that multiple lesions in the biliary tract carry a poor prognosis. The condition of diffuse biliary papillomatosis is characterised by multiple mucus secreting polyps throughout the biliary tree and is probably a distinct clinical entity. An association between diffuse biliary papillomatosis and carcinoma has been postulated.^{13,14} Complete resection of all of the lesions is usually impossible. The frequency of recurrence and the probability of malignant transformation in this condition are likely to produce a poor prognosis.

Two cases have been reported of coexisting biliary tract and intestinal polyps.^{15,16} It has been proposed that bile duct adenomas are part of the spectrum of generalised gastrointestinal polyposis.¹⁷ Some authors have suggested that upper gastrointestinal surveillance of patients with familial adenomatous polyposis should routinely include imaging of the biliary tract.^{15,18}

Benign tumours of the extra hepatic biliary tree are rare. They are prone to recurrence and carry a risk of malignancy. The case of solitary villous adenoma reported here shows that these neoplasms of the biliary tree can be accurately diagnosed by ultrasound guided percutaneous

biopsy in order that the appropriate surgical treatment can be instigated. Surgery is aided by the use of intraoperative ultrasonography.

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