Case report

Adenomyoma of the common hepatic duct

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Background

Adenomyoma occurs most commonly in the fundus of the gallbladder, seldom in other parts of the gallbladder and rarely in the extrahepatic biliary tree, where most lesions are localised to the common bile duct or papilla of Vater. Adenomyoma of the common hepatic duct is extremely rare. To the best of our knowledge, only three cases have been reported so far.

Case outline

A 51-year-old woman was admitted with a three month history of attacks of right upper abdominal pain, nausea, vomiting and fever. Laboratory data, ultrasonography, ERCP and CT confirmed slight cholestasis and proximal bile duct dilatation due to a tumour within the common hepatic duct. Cholecystectomy was performed with excision of the suprapancreatic common bile duct including the convergence of the hepatic ducts plus lymphadenectomy and Roux-en-Y hepaticojejunostomy. Frozen section histology showed the benign nature of the lesion and a tumour-free resection line. Final histology showed adenomyoma. The patient has remained symptomfree for more than 30 months.

Discussion

Although adenomyoma is a benign lesion and the surgical strategy has not been established, complete excision with frozen section is recommended to exclude small malignant foci and local recurrence as well as to avoid surgical overtreatment.

Key words

adenomyoma, common hepatic duct, surgical resection

Introduction

Most tumours of the extrahepatic bile ducts, including the papilla of Vater, are malignant, but benign tumours may occasionally occur. The surgeon should be aware of this fact since surgical over-treatment of benign tumours is inappropriate.

Case report

A 51-year-old woman presented with a three month history of attacks of right upper abdominal pain, nausea, vomiting and fever. She lost 5 kg in weight. On examination, there was nothing remarkable except moderate right upper abdominal tenderness. Blood tests showed an elevated alkaline phosphatase (126 U/L; normal range 30–90 U/L) and gamma glutamyl transpeptidase (60 U/L; normal range <37 U/L). Ultrasonography demonstrated a solid hyperechoic lesion of the common hepatic duct, 50 x 38 mm in diameter, causing dilatation of the proximal bile ducts; the right hepatic duct was 7 mm and the left duct 12 mm in diameter. The gallbladder and distal part of the main bile duct were normal, without stones. Endoscopic retrograde cholangiopancreatography (ERCP) showed a filling defect within the common hepatic duct and dilatation of the proximal biliary tree (Figure 1). Computed tomography (CT) showed a mass in the hilar area of the biliary tree and intrahepatic duct dilatation.

At operation a solid mass in the common hepatic duct was found close to the hepatic duct convergence. As the hepatoduodenal lymph nodes were enlarged, malignancy was strongly suspected. Cholecystectomy was carried with resection of the suprapancreatic part of the common bile duct including the convergence of the hepatic ducts and a lymphadenectomy. The specimen was sent for frozen section, which showed no malignancy and a tumour-free resection margin. The hepatic ducts were joined and then anastomosed to a retrocolic Roux-en-Y jejunal limb. A tiny transjejunal transanastomotic latex tube was inserted into the intrahepatic biliary tree. The recovery was complicated only by wound infection. Tube cholangiogram showed a...
wide hepaticojejunal stoma. The patient remains symptom-free at 30 months.

The resected specimen contained the gallbladder and origin of the common hepatic duct as well as lymph nodes. A large sessile polypoid tumour was found in the common hepatic duct, measuring $45 \times 30 \times 20$ mm, with a broad insertional base ($30 \times 20$ mm) (Figure 2). It was dome-shaped with a smooth, reddish brown surface, firmly attached to common hepatic duct wall, soft and finely granular on the cut surface.

Microscopically, the tumour represented polypoid hamartomatous proliferation composed of multiple branching tubulo-glandular structures irregularly interspersed with muscular fascicles (Figure 3). The glandular epithelium showed no atypia/dysplasia. At its base the tumour was clearly separated from the layers of the common hepatic duct wall, without any sign of (pseudo)invasion. There was an intense stromal inflammatory infiltration, mostly superficial and predominantly eosinophilic, with occasional eosinophilic glandular microabscesses. The regional lymph nodes, including those from the hepatoduodenal ligament, were hyperplastic, with a few reactive epitheloid granulomas but no tumour elements.

**Discussion**

The incidence of benign tumours of the extrahepatic biliary tree is recorded as very low, and they are considered a rarity [1,2]. These tumours include adenomas, papillomatous or polypoid adenomas, adenomyomas and less frequently mesenchimal neoplasms such as neuroma,
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References


leiomyoma, fibroma, fibroadenoma, lipoma, neuroma, melanoma, carcinoid tumour, hamartoma, granular cell tumour and glandular cell myoblastoma [3–7]. Many of these tumours are of mixed pattern with combinations of epithelial, smooth muscle and fibrous tissue. Many tumours of similar histology have been categorised differently by different pathologists [2] which makes it difficult to establish the frequency of the various types [6].

Adenomyoma of the biliary tree is an exceedingly rare lesion, histologically composed of lobules containing ducts and ductules lined by cuboidal or columnar cells and abundant interlacing smooth muscle bundles in the connective tissue stroma [2,4–6]. It is uncertain whether adenomyoma is a true neoplasm or a hyperplastic diverticular and inflammatory lesion, as denoted by the terms adenomyomatous hyperplasia or adenomyomatosis [3–5]. It has been regarded as a slow-growing lesion, although a rapidly-growing variant has also been described [4].

Adenomyoma is rare outside the fundus of the gallbladder, but can arise throughout entire biliary tree including the papilla of Vater. Including the present patient, 32 cases seem to have been reported: 4 in the left duct [2,3,8], 4 in the common hepatic duct [5,7,9], 11 in the common bile duct [3,6,10–18] and 12 at the papilla [4,8,19–25].

Women are three times as commonly affected as men, with a mean age of about 60 years [6]. Adenomyomas of the biliary tree are seldom symptomatic [7], but they can cause some degree of cholestasis with or without cholangitis. There is no specific preoperative investigation except endoscopic biopsy if the tumour arises at the papilla. Frozen section is necessary to avoid radical surgery.

Although the best surgical strategy has not been established, complete excision is recommended to exclude small malignant foci and local recurrence [4–6,9,11], which can be indicated only by frozen section histology of the resection line. However, the lesion itself may require an extensive resection depending upon its exact site, so that biopsy for frozen section may be impossible before such excision has already been performed. We believe that lymphadenectomy should be performed to look for malignant foci since it is relatively simple and safe.