

Traumatic neuroma of the cystic duct with biliary obstruction. Report of a case

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Abstract

A case of amputation neuroma of the biliary tract occurring 12 years after a cholecystectomy is reported. The patient, a 81 year-old man, presented with obstructive jaundice due to a stricture of the extrahepatic biliary tract. The stricture was resected and biliary reconstruction was achieved with a Roux-en-Y jejunal loop. The diagnosis of neuroma was obtained only at histology that showed hyperplastic nerve bundles, positive for protein S 100. The patient is well one year and six months after surgery without signs of recurrence of the stricture. Although the amputation neuroma of the biliary tract has already been reported, it seems worthwhile to emphasise this further report. This lesion is an unusual cause of benign stricture of the biliary tract that may pose difficult diagnostic problems. (*Acta gastroenterol. belg.*, 2003, 66, 28-29).

Key words : traumatic neuroma, benign biliary stricture.

Introduction

Benign strictures of the biliary tract after cholecystectomy are generally due to fibrosis and rarely due to granulomas or amputation neuromas (1). The latter manifest as infiltrating or compressive lesions of the biliary tract resembling a malignancy. We herein, report the clinicopathological features of a case of amputation neuroma of the cystic stump presenting 12 years after cholecystectomy with biliary obstruction. The relative diagnostic and therapeutical implications are discussed.

Case report

A 81 year-old patient was admitted because of obstructive jaundice and pruritus. His past medical history was relevant for blood hypertension, chronic pericarditis and cholecystectomy 12 years before admission. Physical examination showed no abnormalities except for excoriations and jaundice. Laboratory findings showed increased plasma bilirubin (18 mg/dL ; normal range : 0.3-1.0 mg/dL), alkaline phosphatase (857 U/L ; normal range : 39-117), aspartate aminotransferase (189 U/L ; normal range : 10-37 U/L), alanine aminotransferase (153 U/L ; normal range : 10-41 U/L), γ glutamyltransferase (411 U/L ; normal range : 6-50 U/L). Serum levels of tumour markers (Ca 19.9, Carcinoembryonic Antigen and Ca 125) were normal. An abdominal ultrasonographic examination showed dilatation of the intrahepatic bile ducts. A magnetic cholangiopancreatographic (MCP) study showed a focal stricture in the common bile duct (Fig. 1). The patient under-



Fig. 1. — Magnetic resonance cholangio pancreatography showing a stricture in the common bile duct.

went surgery. Laparotomy showed a white-gray, 2 cm large, solid mass, infiltrating circumferentially the main biliary duct. The examination of fresh-frozen sections did not show signs of malignancy. The lesion was resected along with 3 cm of proximal biliary tract and biliary reconstruction was achieved with a Roux-en-Y hepaticojejunostomy. Histology showed hyperplastic nerve bundles, penetrating deeply into the wall of the main biliary duct, mainly composed of Schwann cells, interspersed with collagen fibers and inflammatory infiltrates (Fig. 2). Immunohistochemistry for S 100 protein was positive.

Postoperative course was complicated, at postoperative day four, by the issue of 50 ml of bile from the abdominal drainage. An abdominal ultrasonographic examination showed no collection, and the patient had no symptoms related to the bile leak. Oral feeding was started on postoperative day five. The bile leak decreased progressively and stopped at postoperative day 11. The drainage was removed, and the patient was discharged at postoperative day 14 with no further complication.

One year and six months after surgery the patient is well with no signs of recurrence of the stricture, and his laboratory findings are the following : plasma bilirubin

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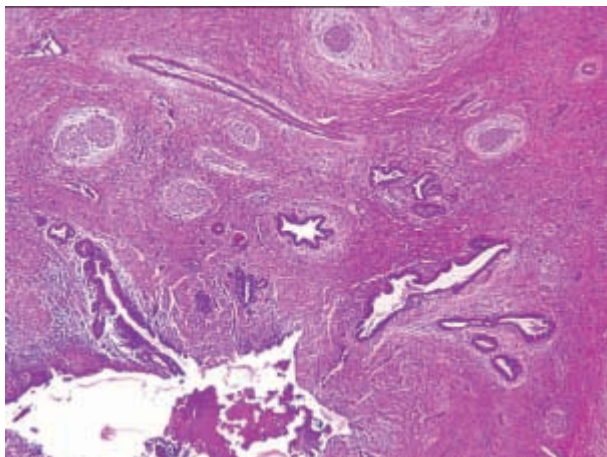


Fig. 2. — Microscopic examination of the operative specimen showed hyperplastic nerve bundles, composed of Schwann cells, interspersed with collagen fibers and inflammatory infiltrates, that penetrate into the wall of the main biliary duct.

0.8 mg/dl (normal range : 0.3-1.0 mg/dL), alkaline phosphatase 211 U/L (normal range : 39-117U/L), aspartate aminotransferase 41 U/L (normal range : 10-37 U/L), alanine aminotransferase U/L 47 (normal range : 10-41 U/L) γ glutamiltransferase 201 U/L (normal range : 6-50 U/L).

Discussion

Neural continuity between the two stumps of a severed nerve is normally ensured by an orderly growth of axons from the proximal to the distal end, occurring through tubes of Schwann cells. Traumatic neuroma occurs when nervous continuity cannot be re-established due to lack of proximity between the two nervous extremities, interposition of granulation and fibrous tissue from adjacent structures, or a missing stump. In this case, axons and Schwann cells at the proximal end of the injured nerve undergo hyperplastic proliferation along with fibroblasts in a dense collagen matrix.

Most of the reports concern the amputation neuroma of the cystic stump after cholecystectomy (1) but it may occur in the main biliary tract after an accidental lesion of the biliary tract (1,2), at the site of a choledotomy, and after liver transplantation (3). Traumatic neuromas are a common pathomorphologic alteration after biliary surgery, as shown by autoptic studies, due to the abundant network of neural tissue surrounding the biliary tract (4-6). After liver transplantation, as many as one quarter of patients may develop a traumatic neuroma (4). They probably originate from the nervous tissue of the recipient, yet are rarely symptomatic. In fact, neural tumours resulting from biliary surgery rarely manifest

clinically, and they are an uncommon cause of stricture of the biliary tract with only about 40 cases reported so far (1). The clinical picture consists of obstructive jaundice due to compression or infiltration of the biliary tract by the neuroma that may extent to the intrahepatic bile ducts (7). Recent advances in non-invasive imaging such as MR cholangiopancreatography may help in defining the extension of the stricture but preoperative differential diagnosis with a malignancy remains difficult. These strictures generally do not respond to non-operative treatment modalities such as percutaneous dilatation or stenting (2,3,8). Surgery is, thus, indicated to establish the diagnosis and to relieve biliary obstruction. This is accomplished through a resection of the extrahepatic biliary tract intimately adhering to the neuroma followed by Roux-en-Y hepaticojejunostomy. In a few cases the intrahepatic extension of the neuroma may render surgical treatment extremely challenging (7).

In the case herein reported, the 12-year long interval of time between the injury and the clinical presentation, the characteristics of the stricture at imaging, and the intraoperative findings suggested a malignancy. As in the majority of the other reported cases, we obtained the diagnosis of amputation neuroma only at histology through the demonstration of nerve bundles positive for S 100 protein.

Although traumatic neuromas are rarely symptomatic, they should be considered in the differential diagnosis of biliary stricture in patients with previous history of biliary surgery.

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