

See discussions, stats, and author profiles for this publication at: <https://www.researchgate.net/publication/51590027>

Post-cholecystectomy amputation neuroma mimicking common bile duct carcinoma

Article in *Acta gastro-enterologica Belgica* · June 2011

Source: PubMed

CITATION

1

READS

227

4 authors, including:



Ivana Mrklić

Klinički Bolnički Centar Split

34 PUBLICATIONS 273 CITATIONS

[SEE PROFILE](#)



Zenon Pogorelić

Klinički Bolnički Centar Split

91 PUBLICATIONS 616 CITATIONS

[SEE PROFILE](#)



Snjezana Tomic

University of Split-School of Medicine, Clinica...

95 PUBLICATIONS 511 CITATIONS

[SEE PROFILE](#)

Some of the authors of this publication are also working on these related projects:



Our experiences with ESIN and ESIN like methods [View project](#)



Molecular cancer diagnostics [View project](#)

All content following this page was uploaded by [Zenon Pogorelić](#) on 21 May 2014.

The user has requested enhancement of the downloaded file.

Post-cholecystectomy amputation neuroma mimicking common bile duct carcinoma

Ivana Mrklič¹, Zenon Pogorelič², Antonia Bendić¹, Snježana Tomić¹

(1) Department of Pathology, Forensic Medicine and Cytology, University Hospital Split and Split University School of Medicine, (2) Department of Pediatric Surgery, University Hospital Split and Split University School of Medicine, Split, Croatia.

To the editor,

Amputation neuromas, also known as traumatic neuromas represent reactive proliferation of nerve fibers that are encased in Schwann cells (1). Amputation neuroma of the common bile duct can occur after cholecystectomy because the common bile duct is surrounded by an abundant nerve supply (2). They always occur at the distal ends of the proximal segments of severed nerves and are characterized by a neural enmeshment in an overgrowth of the Schwannian sheath cells (3). Amputation neuromas are usually found in amputated extremities but they have been reported following radical mastectomy, mastoidectomy, thyroglossal cystectomy and even circumcision (4). Amputation neuromas of the biliary tract are usually asymptomatic, rarely they present with intermittent symptomatic right upper quadrant pain and jaundice. In most cases, the leading differential diagnosis is cholangiocarcinoma due to the similarity of presentation (2,5).

A 65-year-old woman was admitted to the hospital for the investigation of obstructive jaundice. She had a several-month history of continued epigastric pain and jaundice. She had undergone cholecystectomy 22-years ago. On physical examination, she was markedly jaundiced. Her vital signs were within normal limits. Abdomen was soft and painless on palpation. The liver was palpable two fingerbreadths below the right costal margin. The laboratory data included: total bilirubin 37.0 $\mu\text{mol/L}$; direct bilirubin 18.1 $\mu\text{mol/L}$; Aspartate transaminase (AST) 120 U/L; Alanine transaminase (ALT) 127 U/L; Gamma glutamyl transpeptidase (GGT) 299 U/L; alkaline phosphatase 292 U/L. The serum carbohydrate antigen 19-9 (CA 19-9) level was 131 U/ml (normal < 37 U/ml). Other laboratory values were within the normal range. Abdominal ultrasonography demonstrated moderately dilated intra- and extrahepatic ducts. A computed tomography scan of the abdomen revealed a 2.5 cm mass located in the common bile duct (Fig. 1A-B). There was no periportal lymphadenopathy. Endoscopic retrograde cholangiopancreatography revealed a mass in the common bile duct. A biliary stent was placed to relieve the biliary obstruction. Exploratory laparotomy was performed with the presumptive diagnosis of cholangiocarcinoma. At operation many adhesions were found in right upper

quadrant. The common bile duct was dilated and in its middle part there was a firm 2.5 \times 2 cm mass that caused obstruction. Extrahepatic bile duct resection, and Roux-en-Y hepaticojejunostomy reconstruction were performed, following celiac and periportal lymphadenectomy. All margins at surgery were free of malignancy as were the celiac and periportal lymph nodes. Microscopic examination revealed an intramural nodule composed of whorled bundles of nerve fibers surrounded by fibrous scar tissue (Fig. 1C). Immunohistochemically nerve fibers were S-100 and neurofilament positive, and CD34, desmin, actin and CD117 negative. The overlying common bile duct mucosa and the rest of the specimen were without abnormalities.

Cholecystectomy is the most frequently performed operation in abdominal surgery (6). The main goal of both laparoscopic and open cholecystectomy is removal of the gall bladder with minimal risk of injury to surrounding structures (6). Complications of laparoscopic cholecystectomy are infrequent, but include bleeding, infection, pneumonia, blood clots, or heart problems. Neuroma of the biliary tree is very rare complication occurring most commonly in the cystic duct stump after a cholecystectomy and have been described after both laparoscopic and open cholecystectomy (5). The area of the common bile duct bifurcation with the cystic duct is a nerve-rich region and, following a cholecystectomy, the stage is set for the development of an amputation neuroma (2,3,5). Neuroma formation is thought to be precipitated by posttraumatic nerve cell growth after surgery. It is a nonneoplastic disorganized proliferation of axons, Schwann cells, and perineurial cells in a fibrocollagenous stroma and affects nerves that are encased in Schwann cells (1-5). Nerve hypertrophy in response to injury is similar to that occasionally seen with extremity amputations (5). The nerve supply to the gallbladder and its ductal system is composed of sympathetic and parasympathetic nerve fibers, the former through the celiac plexus and the latter through the vagus nerve (5,7). These nerves intermingle to form the anterior and

Correspondence to: Zenon Pogorelič, M.D., Ph.D., Department of Pediatric Surgery, University Hospital Split, Spinčićeva 1, 21 000 Split, Croatia. E-mail: zenon@vip.hr

Submission date: 31/10/2010

Acceptance date: 22/12/2010

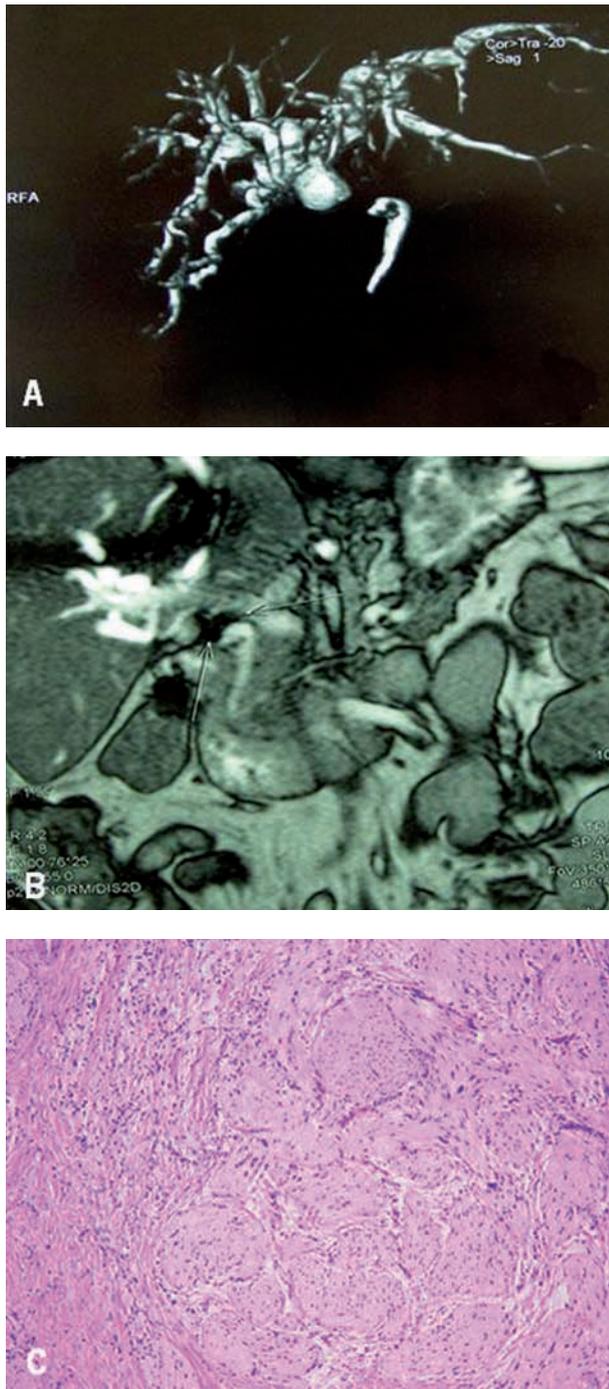


Fig. 1. — (A) A computed tomography scan of the abdomen revealed a tumor in the common bile duct. (B) A space occupying lesion found in the midcommon bile duct. There is bilateral intrahepatic biliary dilation. (C) Nodule composed of whorled bundles of nerve fibers surrounded by fibrous scar tissue H&E, 100 \times .

posterior hepatic plexuses which are located about the region of the union of the cystic and common bile ducts. Stimulation of the sympathetic fibers will primarily produce epigastric pain, whereas vagal stimulation will chiefly bring about dyspepsia and vomiting. The continued epigastric pain in this case might be explained on the

basis of excessive sympathetic stimulation by the tumor. In order to prevent the formation of traumatic neuroma, it is suggested that the nerve trunks being carefully separated mesial to the common bile duct at the junction with the cystic duct.

There are reports in the literature of biliary tree neuroma presenting from several months to 45 years after cholecystectomy (1-3,5). Our patient had a 22-year interval between the initial procedure and the development of symptoms. Most amputation neuromas of the biliary tract occur in the cystic duct stump after cholecystectomy and are asymptomatic. Rarely they present with intermittent or continuous symptomatic right upper quadrant or epigastric pain and jaundice. In one study bile duct neuroma has been demonstrated in up to 10% of post cholecystectomy patients at autopsy (8). However, when they arise in the main hepatic duct and are associated with obstructive jaundice, it is difficult to distinguish them from cholangiocarcinoma. Even if amputation neuroma is suspected in a patient with late-onset jaundice after cholecystectomy, the differential diagnosis from a malignancy is difficult preoperatively.

The tumour marker CA19-9 has been promoted as a reliable test for the detection of pancreatobiliary malignancy, yet its diagnostic role remains poorly defined. Obstructive jaundice is frequently associated with false CA19-9 elevation in benign conditions. Our patient had an elevated CA 19-9 level of 131 U/ml prior to surgery, that is suspected to related to malignancy. Although CA 19-9 levels are often elevated with biliary malignancy such as cholangiocarcinoma, elevated levels are not specific for this diagnosis (5,7,9,10). Marrelli *et al.* studied 128 patients with obstructive jaundice. Elevated CA19-9 levels were found in 61% of benign cases and 86% of malignancies. After biliary drainage, decrease of serum CA19-9 was observed in 19 of 38 malignant cases and in almost all benign cases (10). Mann *et al.* in their study showed that patients with benign conditions generally had modest elevations of CA 19-9 (mean 102 U/ml). In contrast, patients with malignancy generally had much higher CA 19-9 levels (mean 910 U/ml) (9). It is very difficult to discriminate between benign and malignant disease based on the magnitude of CA 19-9 elevation alone (9,10).

The most widely advocated approach in the literature is extrahepatic bile duct resection with clear margins, periportal lymphadenectomy, and Roux en-Y hepatico-jejunostomy (1-3,5,7,8). Unless the diagnosis is definitively known preoperatively, an aggressive resection with formal periportal lymphadenectomy is advocated (5).

The possibility of an amputation neuroma should be kept in mind when patients who underwent cholecystectomy present with pain in the epigastrium, which occasionally radiate to the right side or through the back associated with nausea and sometimes vomiting and in patients with late-onset jaundice after biliary tract surgery. This diagnosis, however, should be made only after an exclusion of the usual causes of these symptoms.

References

1. UENO Y., IKEDA K., MAEHARA M., SAKAIDA N., OMURA N., KUROKAWA H., SAWADA S. Traumatic neuroma of the bile duct. *Abdom. Imaging*, 2008, **33** : 560-562.
2. IANNELLI A., FABIANI P., KARIMDJEE B.S., CONVERSE S., SAINT-PAUL M.C., GUGENHEIM J. Traumatic neuroma of the cystic duct with biliary obstruction. Report of a case. *Acta Gastroenterol. Belg.*, 2003, **66** : 28-29.
3. HOTTA T., KOBAYASHI Y., TANIGUCHI K., NAKA T., JOHATA K., SAHARA M., WATANABE T., TANIMURA H., TSUBOTA Y.T. A traumatic neuroma of the bile duct : a case report. *Hepatogastroenterology*, 2004, **51** : 39-42.
4. TRIGUI D., JALLOULI M., MHIRI R. Penile amputation neuroma after a circumcision. *J. Pediatr. Surg.*, 2008, **43** : 1764.
5. PAQUETTE I.M., SURIAWINATA A.A., ORNVOLD K., GARDNER T.B., AXELROD D.A. Neuroma of the bile duct : a late complication after cholecystectomy. *J. Gastrointest. Surg.*, 2009, **13** : 1517-1519.
6. PERIĆ B., PERKO Z., POGORELIĆ Z., KRALJEVIĆ J. Laparoscopic cholecystectomy in Cantonal Hospital Livno, Bosnia and Herzegovina and University Hospital Center Split, Croatia. *Coll. Antropol.*, 2010, **34** Suppl 1 : 125-128.
7. HYMAN J., WILCZYNSKI S., SCHWARTZ R. Extrahepatic bile duct stricture and elevated CA 19-9 : malignant or benign ? *South Med. J.*, 2003, **96** : 89-92.
8. PICKENS A., VICKERS S., BROWN K., REDDY V.V., THOMPSON J.A. An unusual etiology of biliary hilar obstruction and the potential role of acidic fibroblast growth factor in the development of a biliary neuroma. *Am. Surgeon*, 1999, **65** : 47-51.
9. MANN D., EDWARDS R., HO S. Elevated tumor marker CA 19-9 : clinical interpretation and influence of obstructive jaundice. *Eur. J. Surg. Oncol.*, 2000, **26** : 474-479.
10. MARRELLI D., CARUSO S., PEDRAZZANI C., NERI A., FERNANDES E., MARINI M., PINTO E., ROVIELLO F. CA19-9 serum levels in obstructive jaundice : clinical value in benign and malignant conditions. *Am. J. Surg.*, 2009, **198** : 333-339.