Traumatic Neuroma as a Cause of Obstructive Jaundice

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Abstract

A 70-year-old man with previous cholecystectomy developed progressive obstructive jaundice, 3 months before hospitalization. Intraoperatively, a 2 x 2 x 1.5-cm mass in the distal part of the right hepatic duct was excised to release complete obstruction. It was verified as traumatic (amputation) neuroma. Despite rarity, traumatic neuroma of the bile duct shoud be considered in patients who have antecedent surgical procedure of the biliary tract with subsequent occurrence of extrahepatic biliary obstruction.

Key word : Traumatic Neuroma, Obstructive Jaundice

Extrahepatic cholestasis can result from various causes producing mechanical obstruction of large extrahepatic bile ducts from the porta hepatis to the ampula of Vater(1). Traumatic (amputation) neuroma is one of the rare causes of which the clinical course may mimic malignant condition such as cholangiocarcinoma. We present herewith an amputation neuroma of the distal portion of the right hepatic duct producing obstructive jaundice.

CASE REPORT

A 70-year-old man presented with progressive jaundice for 4-5 months. He had itching skin, pale stool, yellowish urine, and 10 kg loss of body weight. Three years prior to admission, he underwent cholecystectomy because of cholelithiasis. Intraoperatively, there was injury to the common bile duct. Subsequently, he had no jaundice until this episode.

Physical examination showed severe jaundice. The liver span was 13 cm. Laboratory investigation disclosed total birirubin to be 14 mg%, direct birirubin 10 mg%, alkaline phosphatase 2,042 IU/L, SGOT 148 IU/L, SGPT 59 IU/L, and CEA 4.3 ng/ml (normal 0-5 ng/ml). Ultrasonography revealed dilated intrahepatic bile ducts. The common bile duct, how-

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ever, was not clearly demonstrated. No definite mass was detected. The clinical impression was obstructive jaundice due to cholangiocarcinoma.

At exploratomy, a mass causing complete obstruction was found in the distal part of the right hepatic duct. The left hepatic duct was also partially occluded by the lesion. Frozen sections of the resected mass, tissue from the porta hepatis, and suprapyloric lymph node revealed no malignancy. Hepaticojejunostomy and jejunojejunostomy with Roux-en-Y technique were performed afterward.

Pathologic Examination

An unencapsulated, hard, light-brown mass measuring 2 x 2 x 1.5 cm was received. Microscopically, it was composed of numerous nerve fascicles of varying sizes that were arranged haphazardly in hyalinized fibrous tissue (Fig. 1A). The epithelium of bile duct was intervened by these proliferating nerves. Immunohistochemical stainings for neurofilament (NF) and S-100 protein displayed many axons and Schwann cells, respectively (Fig. 1B, 1C). The lesion, therefore, was diagnosed as traumatic neuroma. The tissue from porta hepatis showed severe cholestasis. The lymph node revealed reactive hyperplasia. Postoperatively, jaundice gradually decreased. The patient became healthy and had no jaundice in the latest follow-up, 4 months after surgical intervention.

DISCUSSION

The common differential diagnoses of extrahepatic obstructive cholestasis include lithiasis, malignant tumor of the bile duct and periampullary

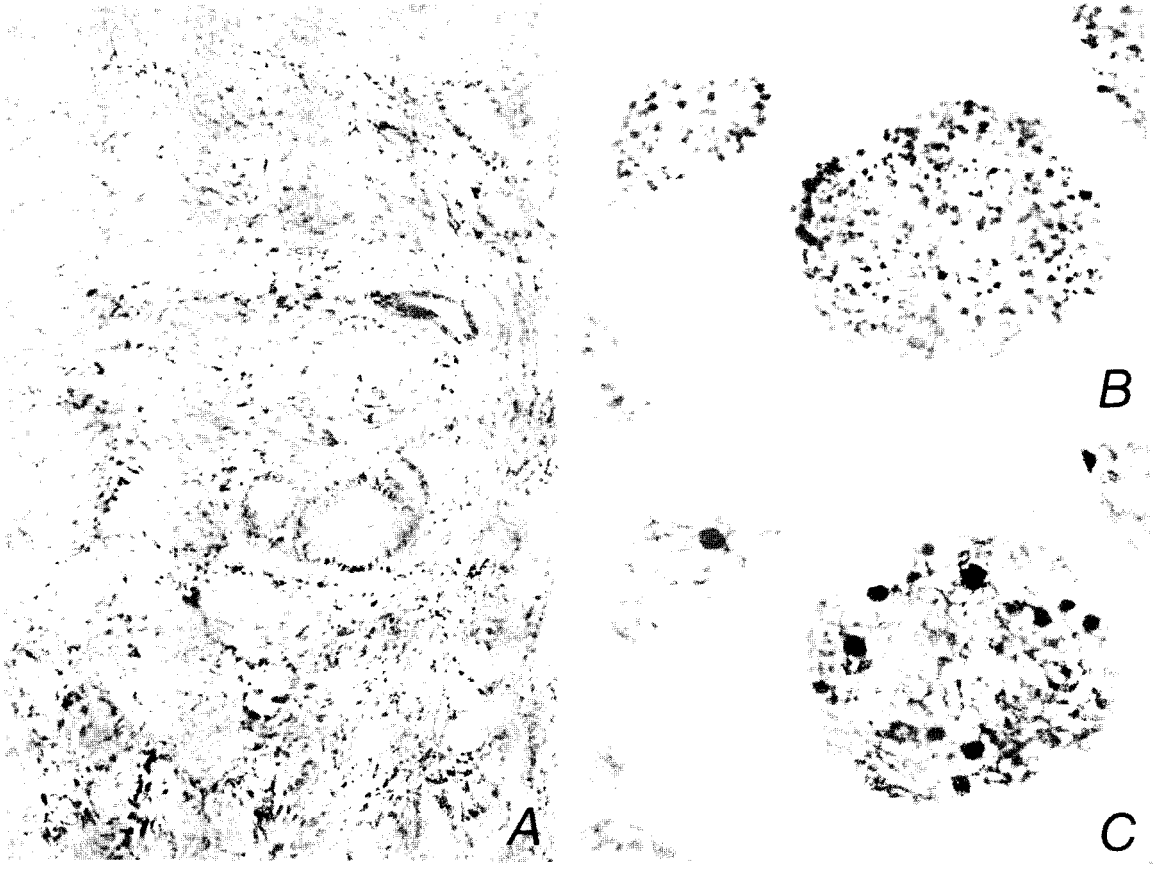

Fig. 1. A: Numerous nerve fascicles infiltrate in the lamina propria of the right hepatic bile duct in disorderly fasion and are intervened by collagenous stroma. Residual bile duct epithelium is seen. B: Multiple neurofilament-positive axons are seen as dark spots in cross section. C: Same microscopic field as B. S-100 protein positivity is noted in cytoplasm of cells interpreted as Schwann cells which forms circular sheaths around the axons.

region, and metastasis of lymph nodes at porta hepatis[1]. In patients with previous cholecystectomy, stricture or stenosis of the bile ducts, traumatic neuroma, and biliary pseudocyst should be considered[2]. The stricture of bile duct mostly presents soon after the operation[3]. In cases of traumatic neuroma, the onset of jaundice may occur as early as 6 months or as late as 35 years with a mean duration of 8 years[4]. The gross appearance and location of traumatic neuroma may be similar to those of cholangiocarcinoma[5,6]. Therefore, intraoperative frozen section should be done in order to design an appropriate surgical procedure.

The formation of truamatic neuroma is related to unaccomplishment of reinnervation of the injured nerves ; thus, it is not a true neoplasm. The biliary system is susceptible for development of traumatic neuroma because there are abundant sympatatic nerve plexuses in this area[5]. Most traumatic neuromas are asymptomatic and may be found microscopically after cholecystectomy with an incidence of 18-27 per cent on postmortem examinations[4]. After liver transplantation, traumatic neuromas have been reported in 26 of 93 cases (27.9%). Most of which (25 cases) are asymptomatic[7]. However, in the absence of previous biliary tract surgery, encapsulated spontaneous neuroma and typical traumatic neuroma have also been reported[8,9]. Microscopically, spontaneous neuroma is encapsulated and the proliferating axons take place in the endoneurial space within the confines of the perineurium[8].

Grossly, traumatic neuroma may appear as a whitish and firm fusiform mass which can be intra- or extraluminal. Extension of traumatic neuroma upward, even into intrahepatic bile duct, or downward is probable[4,10-13]. Common hepatic duct is the most common location of traumatic neuroma causing obstruction[5].

Histologically, traumatic neuroma consists of a poorly defined and disorderly growth of nerve fascicles. Each contains all usual components of nerve including axons, Schwann cells, fibroblasts and perineurial cells, and is surrounded by dense fibrosis[14,15]. However, traumatic neuroma must be distinguished from such conditions as mucosal neuroma, plexiform neurofibroma, and plexiform neurilemmoma.

Mucosal neuroma is almost always multiple and associated with the multiple endocrine neoplasia syndrome type II B[14]. Microscopically, it may superficially resemble traumatic neuroma. A distinction can be made by the presence of loose intervening myxoid stroma in the mucosal neuroma[15].

Plexiform neurofibroma, a pathognomonic feature of type I von Recklinghausen's disease, must also be distinguished from traumatic neuroma[14, 16]. The presence of myxoid change suggests the possibility of the former[14]. In addition, individual axons in neurofibroma are separated by the neoplastic Schwann cells, fibroblasts, and matrix rich in mucopolysaccharide and collagen[16]. This finding is helpful for differentiation of plexiform neurofibroma, either from traumatic neuroma or from plexiform neurilemmoma, in which neuroma contains normal components of the nerve fascicle and the other is composed mainly of Anthoni type A of neoplastic Schwann cells[14,16].

Immunohistochemical study in our case demonstrated NF-positive nerve fibers, and S-100 protein-positive Schwann cells in each nerve fascicle supporting the diagnosis of traumatic neuroma.

Inspite of infrequent occurrence, traumatic neuroma should be considered in patients who have obstructive jaundice especially after previous surgical intervention of the biliary system.

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ดีซ่านที่เกิดจาก ทรอมาติก นิวโรมา อุดกั้นท่อน้ำดี

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ผู้รายงานได้นำเสนอผู้ป่วยชายไทย อายุ 70 ปี 3 ปีก่อนมาโรงพยาบาลเคยได้รับการผ่าตัดถุงน้ำดีเนื่องจากตรวจพบนิ่ว ครั้งนี้มาพบแพทย์ด้วยอาการตัวเหลืองอย่างค่อยเป็นค่อยไปในเวลา 3 เดือน การทำผ่าตัดพบก้อนขนาด 2 x 2 x 1.5 ซม ที่ท่อน้ำดี ผลการตรวจทางพยาธิวิทยาพบเป็น ทรอมาติก นิวโรมา

ดีซ่านที่เกิดจากการอุดตันของท่อน้ำดีมีสาเหตุได้หลายประการ ทรอมาติก นิวโรมา เป็นสาเหตุหนึ่งที่พบได้น้อย แต่ควรจะต้องนึกถึงเสมอโดยเฉพาะในผู้ป่วยที่มีประวัติการทำผ่าตัดบริเวณนี้มาก่อน การทำ frozen section สามารถช่วยในการวินิจฉัยแยกกับรอยโรคมะเร็งได้

คำสำคัญ : ทรอมาติก นิวโรมา, ดีซ่าน

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