

Case Report

Traumatic Neuroma Arising from Surgical Trauma during Conversion from Laparoscopic to Open Cholecystectomy

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Traumatic neuroma is an abnormal proliferation of injured nerves resulting from trauma or surgery. We present a case of traumatic neuroma arising in the cystic duct after cholecystectomy. A 66-year-old man was referred to our department due to a biliary tumor. He had undergone cholecystectomy 20 years prior. Cholangioscopy showed an elevated lesion covered with smooth mucosa. Histological examination revealed normal bile duct mucosa. Although benign disease was suspected, the possibilities of malignant disease could not be excluded. Extrahepatic bile duct resection was planned to include intraoperative rapid-freezing of a biopsy specimen followed by histopathological examination. These intraoperative histology results showed proliferation of nerve and fibrous tissue only, resulting in the diagnosis of traumatic neuroma, so no lymph nodes were removed. To avoid excessive surgical intervention, histopathological examination of an intraoperative rapid-frozen biopsy specimen may be important for diagnosing traumatic neuroma.

Key words: traumatic neuroma, biliary stricture, cholecystectomy, cholangiography, intraoperative rapid-frozen biopsy

Traumatic neuroma is a rare, non-neoplastic lesion that forms at the proximal end of a damaged nerve following trauma or surgery as a result of a healing process involving the hyperplastic proliferation of nerve fibers and connective tissue [1]. The lesion can develop at any time after surgery. Traumatic neuroma arises more frequently in the lower limbs, followed by the head and neck [2]; it has also been reported to develop within the biliary tract as an unintended consequence of surgical trauma experienced during cholecystectomy [3]. Traumatic neuroma may mimic malignant tumors due to the vague clinical and imaging features it displays. Herein, we present a case of traumatic neuroma arising in the cystic duct after cholecystectomy

and describe its clinical features including those that sometimes make intraoperative rapid-frozen sectioning necessary.

Case Presentation

A 66-year-old man was referred to our department because of an abnormality detected during abdominal ultrasonography. Two years prior, he had undergone cholecystectomy that had required conversion from laparoscopic to open surgery. Otherwise, his medical history included essential hypertension, diabetes mellitus, and dyslipidemia. He also underwent percutaneous intervention for ischemic heart disease, and pacemaker implantation due to a complete atrioventricular

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block. He had been prescribed aspirin, some hypertensive drugs, statin, and an oral hypoglycemic agent. Laboratory investigation showed no abnormality. Circulating liver enzymes and serum bilirubin were within normal limits. Carbohydrate antigen 19-9 (21.9 U/mL) and carcinoembryonic antigen (1.5 ng/mL) were not elevated. An abdominal contrast-enhanced computed tomography (CT) scan showed a 10-mm enhanced nodule in the extrahepatic bile duct (Fig. 1). Endoscopic retrograde cholangiopancreatography (ERCP) revealed a hemispherical defect in the bile duct; however, biliary dilation was not detected (Fig. 2A). Cholangioscopy revealed an elevated lesion covered with smooth mucosa (Fig. 2B), and a biopsy of this elevated portion showed normal bile duct mucosa with no evidence of malignancy. Endoscopic ultrasonography (EUS) had previously revealed the presence of an 8.4-mm hypoechoic nodule (Fig. 2C).

The possibility of a cholangiocarcinoma arising from the extrahepatic bile duct was not supported by the findings of cholangioscopy or its biopsy. Although benign disease including traumatic neuroma were suspected, the possibilities of malignant disease arising from gallbladder cancer in the remnant cystic duct or a neuroendocrine tumor could not be excluded. Since the lesion was asymptomatic, the patient was offered two options: either follow-up at a later time or plan a surgical resection. The patient opted for surgical treatment, and a surgical intervention was planned accordingly after the surgical indications had been discussed in detail.

First, adhesiolysis of the upper abdominal cavity was done. The palpable mass was found near the remnant cystic duct. The mass adhered robustly to the sur-

rounding tissues, and the common bile duct could not be separated from the mass. Therefore, extrahepatic bile duct excision was carried out. A histopathological examination of the intraoperative rapid frozen section biopsy revealed proliferation of nerve and fibrous tissue with no cancer cells. Specifically, the histological examination revealed an irregular proliferation of nerve bundles (Fig. 3A, B). Immunohistological examination revealed that the nerve bundles were composed of axons that were positive for neurofilament staining (Fig. 3C) and Schwann cells positive for S-100 protein staining (Fig. 3D). The patient was finally diagnosed with traumatic neuroma due to his history of cholecystectomy. Hence, the surgery did not proceed to lymphadenectomy. Instead, Roux-en Y hepaticojejunostomy was carried out as part of the biliary reconstruction. The postoperative course of the patient was uneventful. The patient was discharged on postoperative day 10 without any complications.

Discussion

Traumatic neuroma can develop in the biliary tract. Cholecystectomy and liver transplant surgical interventions are a major cause of biliary traumatic neuroma [3]. Open surgeries such as cholecystectomy and liver transplantation are conducted frequently, while laparoscopic cholecystectomy is relatively rare [3]. Wang *et al.* included 88 biliary traumatic neuromas in his review of traumatic neuromas from abdominal surgery [4]. We also searched PubMed for reports of traumatic neuroma among the English-language literature. We created a table of patient characteristics based on Wang's review

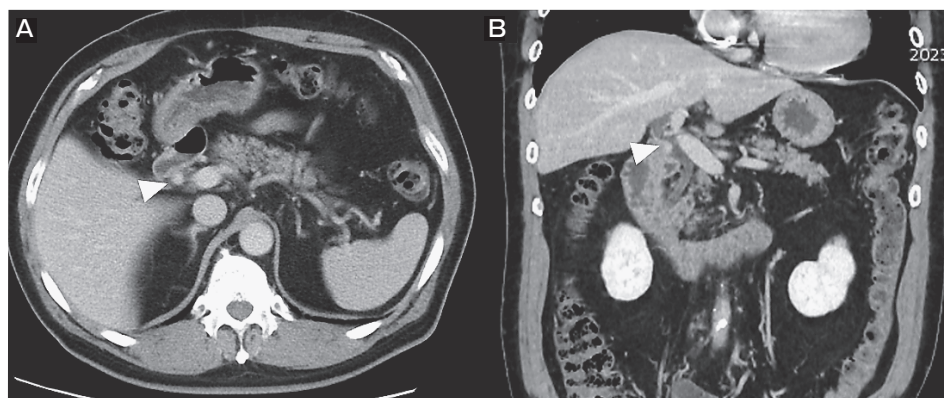


Fig. 1 Contrast-enhanced computed tomography showed an enhanced nodule in the extrahepatic bile duct (arrowhead).

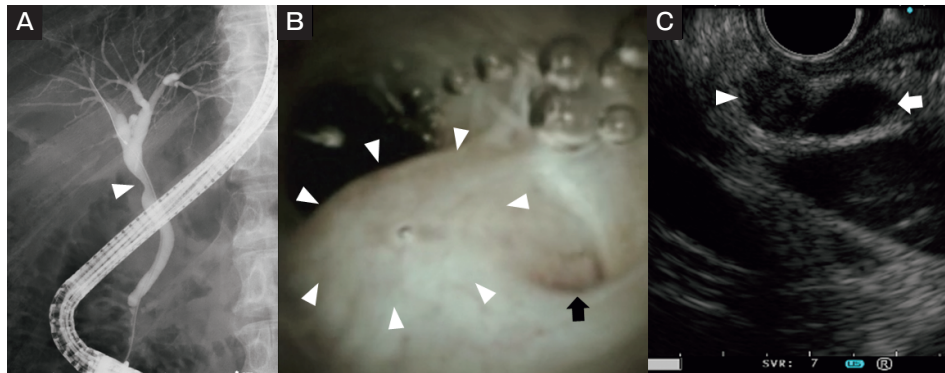


Fig. 2 Cholangiopancreatography, cholangioscopy, and endoscopic ultrasonography results. **A**, Endoscopic retrograde cholangiopancreatography revealed a hemispherical defect of the bile duct (arrowhead); **B**, Cholangioscopy revealed an elevated lesion covered with smooth mucosa (arrowhead) near the aperture of the cystic duct (arrow); **C**, Endoscopic ultrasonography (EUS) revealed the presence of an 8.4-mm hypoechoic nodule (arrowhead) adjacent to the bile duct (arrow).

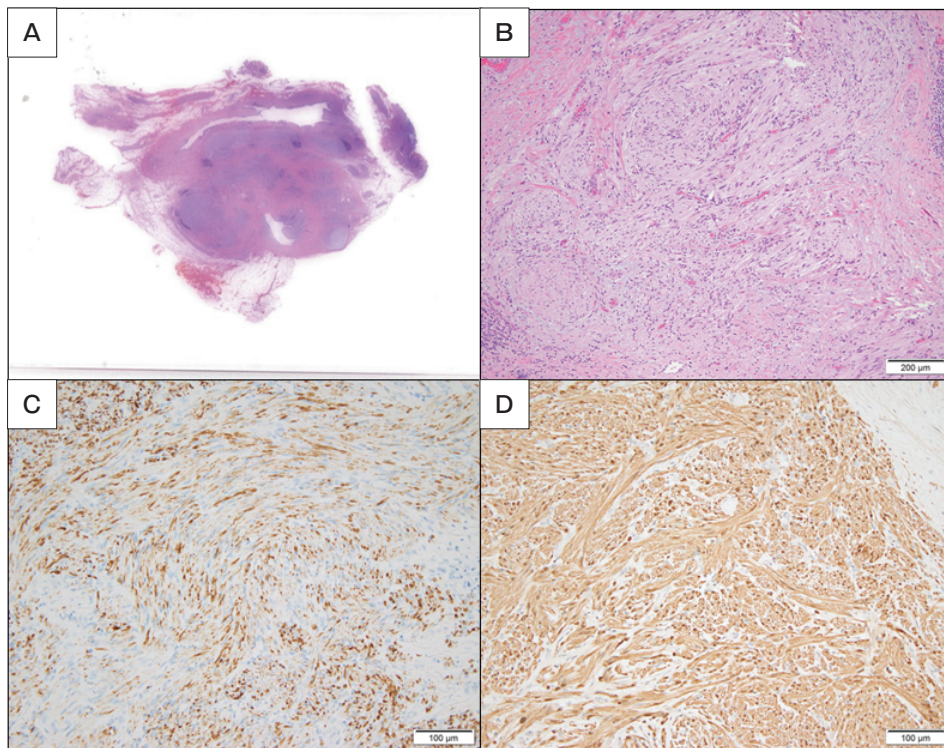


Fig. 3 Histological and immunohistological findings. **A**, Loupe image of the specimen; **B**, Hematoxylin and eosin staining revealed irregular proliferation of nerve bundles; **CD**, Immunohistological examination confirmed that the nerve bundles were positive for neurofilament staining (b) and S-100 protein staining.

[4], recent case reports [5-9], and our case (Table 1). The incidence was higher in males than females. The median age at diagnosis of traumatic neuroma after biliary surgery was 62 years, with nearly 70% of patients

being over 60 years old. The interval between initial surgery and the diagnosis varied from 1 month to 56 years. The interval was generally longer after biliary surgery than after liver transplantation. Since traumatic

Table 1 Patients information and treatment option for the patient with traumatic neuroma around the bile duct

	After biliary surgery n = 38		After liver transplantation n = 30	
Age, years old, medeian (range)	62 (33–81)		54 (16–65)	
Sex, male, (%)	26 (70)		20 (80)	
First operation, (%)				
	OC	26 (68)		
	LC	8 (21)		
	Hepatectomy	3 (8)		
	Cholecystectomy*	1 (3)		
Clinical presentation, (%)				
	Jaundice	19 (50)	Jaundice	20 (67)
	Abdominal symptoms**	10 (26)	Increased liver enzymes	5 (17)
	Increased liver enzymes	3 (8)	Cholangitis	3 (10)
	none	6 (16)	Bile leakage	1 (3)
			Abdominal discomfort	1 (3)
Interval, medeian (range)	12 years (1 month–56 years)		10 months (1 months– 19 years)	
Treatment, (%)				
	BDR	25 (66)	Resection	20 (67)
	Local resection	5 (13)	re-liver transplantation	10 (33)
	PD	4 (11)		
	Hepatectomy	2 (5)		
	Observation	2 (5)		

OC, open cholecystectomy; LC, laparoscopic cholecystectomy; BDR, extrahepatic bile duct resection; PD, pancreaticoduodenectomy.

*, no information about open or laparoscopic; **, abdominal pain or discomfort.

neuroma is often painful, some patients complained of right upper quadrant pain [10]. However, the majority of patients were asymptomatic unless biliary strictures arose [3]. In the current case, the biliary mass was detected incidentally by CT scan. Wang *et al.* also reviewed traumatic neuroma due to other abdominal surgeries [4]. It is worth keeping in mind that traumatic neuroma may occur after any surgery in the abdomen.

In adults, strictures may arise in the extra-hepatic bile duct due to either malignant or benign biliary tumors. Postoperative complications include anastomotic strictures or iatrogenic injury and are secondary to chronic disease such as primary sclerosing or ascending cholangitis [11]. The most common type of primary biliary tumor is cholangiocarcinoma; benign tumors occur much less frequently, accounting for only 5.2% of all biliary tumors [12]. Therefore, once a biliary tumor has been documented, surgical management should be considered.

Preoperative diagnosis is the most difficult aspect of the management of biliary traumatic neuroma. A previous report found no specific imaging features for traumatic neuromas of the bile duct [8]. Generally, the most common ultrasonographic imaging findings for traumatic neuromas is a hypoechoic lesion [7,8]. Contrast-enhanced CT generally reveals homogeneously enhanced nodules [8,9]. Cholangiography including ERCP, magnetic resonance cholangiopancreatography (MRCP) or via percutaneous transhepatic biliary drainage (PTBD) typically shows a smooth stricture of the bile duct with or without biliary dilation [3,7,13]. Cholangioscopy may represent the most important tool for diagnosing biliary traumatic neuroma. To our knowledge, only two prior reports in the English-language biomedical literature have described cholangioscopic findings [14,15]. According to these reports, a smooth elevated lesion covered with normal mucosa was detected [14,15]. In the present case, CT

revealed the presence of an enhanced nodule around the extrahepatic bile duct. MRCP and ERCP showed a unilateral smooth stricture of the bile duct. Peroral cholangioscopy revealed an elevated lesion of the bile duct near the confluence of the cystic duct. We suspected benign disease because of the smooth surface of the lesion. Although a biopsy of the lesion obtained via ERCP showed normal mucosa of the bile duct, we could not rule out the occurrence of malignancy.

Surgical resection is commonly indicated for traumatic neuroma because the lesion may be cholangiocarcinoma or because its benign versus malignant nature cannot be confirmed preoperatively [8]. However, traumatic neuroma is not malignant disease. Therefore, once traumatic neuroma has been diagnosed, continued observation of the neuroma is permitted in an asymptomatic situation. Although biopsy is extremely important for diagnosis, biopsy of biliary tumors is challenging. Only two cases of preoperative traumatic neuroma diagnosis have been reported. One case was diagnosed using EUS-guided biopsy [14]; the other was diagnosed using biliary biopsy via peroral cholangioscopy [15]. Recently, a few reports have described EUS-guided Fine Needle Aspiration (EUS-FNA) as being superior to ERCP tissue sampling for suspected malignant biliary lesions [16,17]. Since traumatic neuroma generally develops outside of the bile duct, EUS-FNA seems to be a more useful tissue sampling procedure for traumatic neuroma than for malignant biliary disease. In the current case, surgical resection was indicated because bile duct cancer could not be excluded due to the difficulty of tissue sampling. However, EUS-FNA has the potential to provide a true diagnosis preoperatively despite its small sample size.

Traumatic neuroma is generally a candidate for surgical resection due to suspicion of malignant disease. Extended surgical resection including lymphadenectomy is sometimes performed [17]. However, the possibility of diagnosing suspected neuroma based on intraoperative histology of rapid-frozen specimens has been demonstrated [9]; this practice spares the lymph nodes. In the current case, we suspected traumatic neuroma preoperatively. However, we could not confirm the benign nature of the lesion due to the difficulty of obtaining a biopsy. We finally decided upon surgical resection after meticulous discussion. We ultimately performed extra hepatic bile duct resection and Roux-en Y hepaticojejunostomy without lymphadenec-

tomy because the intraoperative frozen section diagnosis revealed proliferation of nerve fibers but no malignant cells. When traumatic neuroma is suspected before the operation, surgeons should conduct an intraoperative rapid-frozen section biopsy diagnosis to avoid excessive surgical invasion for the patient.

In conclusion, distinguishing malignant disease from traumatic neuroma can be challenging. To avoid excessive surgery, histopathological examination of an intraoperative rapid-frozen biopsy specimen can be important for diagnosing traumatic neuroma.

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