Case report

# Traumatic Biliary Neuroma Mimicking a Malignant Tumor

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**Neuroma traumático de la vía biliar simulando una neoplasia maligna**

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No conflicts of interest.



**Abstract**
**Case presentation.** A 53-year-old woman presented abdominal pain, weight loss, fever, choluria, and conjunctival jaundice. During the physical examination, the eyes were observed with icteric staining 3+/4+, and the abdomen had abundant adipose panniculus without signs of peritoneal irritation. Patient with history of a laparoscopic cholecystectomy 12 year ago. **Treatment.** An endoscopic retrograde cholangiopancreatography was performed, which showed an impassable critical stenosis in the main biliary tract. Computed tomography showed dilatation of the intrahepatic bile duct. Blood tests showed a cholestatic pattern with elevation of the tumor marker CA19-9. At a multidisciplinary conference, it was decided to opt for a surgical approach, and a resection of the extrahepatic bile duct with lymph node dissection and reconstruction with the Abdo-Machado technique was performed. The pathological report was compatible with a traumatic neuroma of the biliary tract. **Outcome.** She presented a Clavien-Dindo IVa type complication, was treated in the Critical Care Unit with favorable evolution, resolving the complication, and was discharged in stable condition from the General Surgery Service. At six months postoperatively, she has not presented any incident.

**Keywords**
Neuroma, Bile Ducts, Neoplasms.

**Resumen**
**Presentación del caso.** Una mujer de 53 años que consultó de emergencia por un cuadro de dolor abdominal, pérdida de peso, fiebre, coluria e ictericia en las conjuntivas. Durante el examen físico se observaron los ojos con tinte ictérico 3+/4+, y el abdomen con abundante panículo adiposo sin signos de irritación peritoneal. Paciente con antecedente de una colecistectomía laparoscópica de hace 12 años. **Intervención terapéutica.** Se le realizó una colangiopancreatografía retrógrada endoscópica que evidenció una estenosis crítica no franqueable en la vía biliar principal. La tomografía axial computarizada mostró una dilatación de la vía biliar intrahepática. Los exámenes de sangre mostraron un patrón colestásico con elevación del marcador tumoral CA19-9. En una conferencia multidisciplinaria se decidió optar por un abordaje quirúrgico y se realizó una resección de la vía biliar extrahepática con disección ganglionar y reconstrucción con técnica de Abdo-Machado. El reporte patológico fue compatible con un neuroma traumático de la vía biliar. **Evolución clínica.** Presentó una complicación de tipo Clavien-Dindo IVa, fue atendida en la Unidad de Cuidados Críticos con evolución favorable, resolviendo la complicación y fue dada de alta en condición estable del Servicio de Cirugía General. A los seis meses posoperatorios, no ha presentado ningún incidente.

**Palabras clave**
Neuroma, Conductos Biliares, Neoplasia.

## Introduction

Bile duct neuromas are rare diseases. Most extrahepatic bile duct lesions are malignant, with only 6 % being benign in origin.[i] In the absence of neurofibromatosis, these neuromas are associated with trauma or surgical manipulation of the biliary tract.[ii] Traumatic neuromas are non-neoplastic lesions that form at the proximal end of the injured nerve as a result of a scarring process and hyperplastic proliferation of nerve fibers and connective tissue.[iii] Most of the reported cases follow cholecystectomy.[ii]

Symptoms usually appear many years after the surgical procedure. Traumatic neuromas originating from the cystic duct remnant following cholecystectomy are rare. The

cases that have been published are related to open cholecystectomy and only a limited number to laparoscopic cholecystectomies.[iv,v] These neuromas are usually located in the cystic duct stump.[vi] This report describes the case of a patient with a traumatic bile duct neuroma, including its different diagnostic, therapeutic, and prognostic implications due to the low incidence of the disease that simulated a malignant neoplasm.

## Case presentation

A 53-year-old woman consulted the emergency department for a one-day clinical picture of diffuse abdominal cramping abdominal pain, not associated with food intake, nausea, vomiting, fever, choluria, and conjunctival jaundice; in addition, she presented unquantified and unintentional weight loss of two months of evolution. Vital signs were reported without abnormalities: blood pressure of 120/80 mmHg, heart rate of 86 bpm, temperature of 37. 5 °C, weight of 104 kg, and height of 1. 65 m, with a body mass index (BMI) of 38. 2 kg/m$^2$. Physical examination revealed eyes with icteric staining 3+/4+ and an abdomen with abundant adipose panniculus, normal peristalsis, and no signs of peritoneal irritation.

Among the relevant history, she underwent laparoscopic cholecystectomy 12 years ago for cholelithiasis, with no apparent complications at discharge. Laboratory tests showed leukocytosis and cholestasis (Table 1). An abdominal ultrasound was performed, which showed dilatation of the extrahepatic and intrahepatic bile duct, choledochus of 0. 9 cm, right hepatic radicle of 0. 81 cm, and left hepatic radicle of 0. 9 cm, without any other relevant findings.

## Treatment

The patient was evaluated by the hepatopancreatic-biliary surgery team of the institution, which diagnosed acute cholangitis grade II (according to Tokyo criteria 13/18). She presented morbid obesity, accompanied by an icteric-cholestatic syndrome. An endoscopic retrograde cholangiopancreatography (ERCP) was performed, which reported the extraction of a common bile duct stone, a critical hourglass stenosis in the common hepatic duct that did not allow the guidewire to advance, with dilatation of the intrahepatic bile duct and outflow of clear biliary material (Figure 1). Due to the cholangitis, the infectious disease team indicated the use of piperaziline/ tazobactan 4.5 g every six hours, and antipyretics; also, they recommended the control of the septic focus as soon as possible. The patient presented a satisfactory evolution, took antibiotics for seven days, and showed an improvement in the inflammatory and cholestatic pattern.

The patient presented a satisfactory evolution, took antibiotics for seven days, and showed an improvement in the inflammatory and cholestatic pattern. Due to the endoscopic findings of ERCP and the elevation of the tumor marker (CA19-9), the possibility of a malignant neoplasm of the extrahepatic biliary tract (Klatskin tumor type I) was suspected. An abdominopelvic CT scan was performed for staging, which did not demonstrate any findings suggestive of locally advanced or distant disease. The patient was evaluated in a multidisciplinary conference, and consensus was reached that she was a candidate for surgical resection due to the suspicion of malignancy and the need to resolve the icteric picture. Ten days after her admission and having completed treatment for cholangitis, an exploratory laparotomy was performed with lymph node emptying of the hepatoduodenal ligament, resection of the main biliary tract, and an Abdo-Machado type biliodigestive bypass. Due to the suspicion of malignancy, freeze biopsies of the proximal and distal edges of the bile duct were sent, which were negative for malignancy. The estimated transoperative blood loss was approximately 300 mL, with an operative time of 270 minutes.

## Outcome

The postoperative process was managed with the "fast-track" strategy, which consists of a set of strategies for accelerated postoperative recovery, including early initiation of enteral feeding and early ambulation. The patient had a favorable evolution and only required analgesia (paracetamol 1g intravenous every six hours) and was discharged seven days after surgery. However, ten days after hospital discharge, the patient consulted again in the emergency unit due to abdominal pain and unquantified fever. The vital signs recorded in the emergency unit were: blood pressure of 100/60 mmHg, heart rate of 120 bpm, respiratory rate of 24 rpm, and temperature of 38. 5 °C. Laboratory tests showed a severe infectious process (Table 2). He presented signs and symptoms of dyspnea and diffuse abdominal pain, with the operative wound clean and without abnormalities. For this reason, an abdomino-pelvic computed axial tomography was performed, which reported a fluid collection in subphrenic and perihepatic space of large volume (Figure 2).

An interventional radiologist percutaneously drained this collection with a multipurpose pigtail drain, extracting 350 mL of purulent fluid, which was sent for bacteriological culture (Figure 2).

During the second admission, the patient's condition was classified as septic shock secondary to an infectious abdomen, which required management in the Intensive Care Unit with vasopressor support (noradrenaline at response dose) and adequate management of acute renal failure. Carbapenemase-producing *Escherichia coli* and *Pseudomona aeuruginosa* were isolated from the abdominal collection. The patient was treated with tigecycline 50 mg intravenous every 12 hours, amikacin 1 g intravenous every day, plus polymyxin B 500 000 IU intravenously every 12 hours for 10 days. Because the patient presented multiorgan dysfunction and required intensive care unit support, the condition was classified as Clavien-Dindo IVa, a standardized way of classifying postsurgical complications based on their therapeutic implications. Fifteen days after drainage of the collection, the CT scan was repeated, which showed no evidence of persistence of the collection, so the percutaneous drainage catheter was removed, and the patient was discharged from the hospital with no further complications. At six months of follow-up, the patient has not presented any eventuality.

**Table 1.** Laboratory tests

| Test | Results |
|---|---|
| Hemoglobin | 13 g/dL (NV: 12-16) |
| White blood cells | 12 200 (NV: $10^3$/µL) |
| Neutrophils | 73 % (37-70) |
| Total bilirrubin | 8.15 mg/dL (NV up to 1.1 mg/dL) |
| Direct bilirrubina | 5.91 mg/dL |
| Alkaline phosphatase | 272 U/L (NV: 35-104) |
| AST | 97 U/L (NV: 0.0-35) |
| ALT | 141 U/L (NV: 0.0-35) |
| C-reactive Protein | 1.59 mg/dL (NV: 0.0-0.5) |
| CA19-9 | 287 U/mL (NV: 0.0-39) |

NV: Normal value. AST: Aspartate aminotransferase. ALT: Alanine aminotransferase.
Source: Clinical laboratory data from the ISSS Medical, Surgical and Oncological Hospital.

## Clinical diagnosis

The proximal and distal edges of the bile duct were sent for transoperative pathological analysis for the diagnostic suspicion of an extrahepatic cholangiocarcinoma, and the results were negative for malignancy.

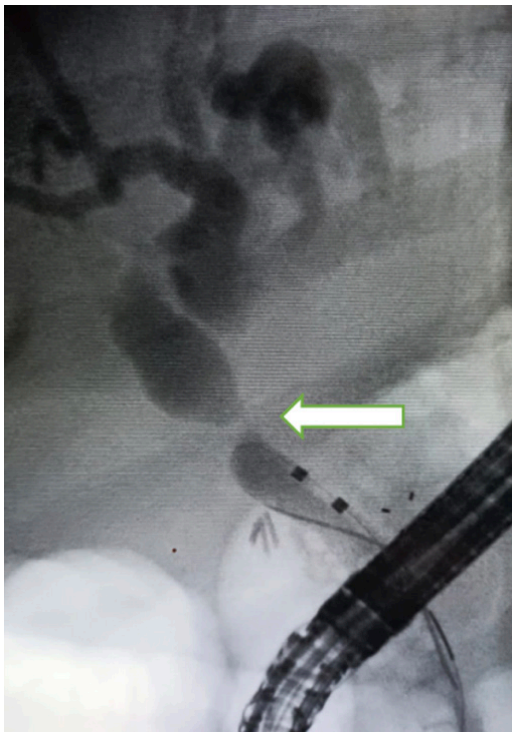

**Figure 1.** Endoscopic retrograde cholangiopancreatography (ERCP) shows a critical hourglass stenosis at the level of the main bile duct (arrow) that simulates a neoplasm and dilatation of the bile duct

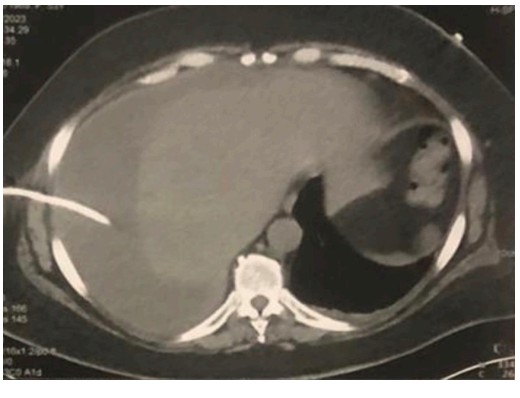

**Figure 2.** Computed axial tomography of the abdomen. Subphrenic and perihepatic collection of approximately 300 mL with a drainage catheter placed inside

The Pathology Service team evaluated the samples and concluded that it was a neuroma of the biliary tract, composed of a firm, whitish nodule measuring 0. 9 cm x 0. 8 cm, corresponding to a benign mixed neoplasm consisting of fibroblastic spindle cells organized in a lobular pattern, with no evidence of malignancy on examination (Figure 3). By microscopy, the characteristic arrangement of this disease could be observed, with positive labeling for CD34 for fibroblastic cells (Figure 4A and 4B). In addition, immunohistochemistry showed positivity for S100 and a mitotic index (ki67) of

two. (Figure 5). A diagnosis of traumatic biliary neuroma was made due to the surgical history and pathologic findings.

## Discussion

Bile duct neuroma was first reported in 1928;[vi] however, most published information on the subject consists of individual case reports or case series. In the absence of neurofibromatosis, neuromas are associated with trauma or surgical manipulation of the bile duct.[ii] Traumatic neuromas are non-neoplastic lesions that form at the proximal end of the injured nerve, secondary to a scarring process and hyperplastic proliferation of nerve fibers and connective tissue.[iii] Traumatic injuries to peripheral nerves lead to multimodal cell proliferation, regeneration failure, and deformation of the nerve structure.[vii] These lesions are generally associated with surgery, especially amputations, with the extremities being the most frequent sites of localization.[iii]

Traumatic neuromas of the digestive system are rare, and generally when they occur in the biliary tract, they follow cholecystectomy or liver transplantation.[viii] The patient has a history of cholecystectomy, one of the main causes of the disease. In most documented cases, neuromas occurred after open cholecystectomy and only sporadically are secondary to laparoscopic surgery.[ix] The incidence is higher in men, although women more frequently develop cholelithiasis.[x] Furthermore, the frequency of traumatic neuromas of the biliary tract increases with age, with more than 70 % of cases occurring in those over 60 years of age.[viii] Some case reports show that the interval of presentation between surgery and diagnosis of neuroma is between two months to 46 years;[viii,xi] in this case, the interval was 12 years. Approximately 65 % of

**Table 2.** Hospital readmissions laboratory tests

| Test | Results |
|---|---|
| White blood cells | 18 100 (NV: $10^3$/µL) |
| Neutrophils | 87 % (37-70) |
| Creatinine | 5.7 mg/dL (NV: 0.5-0.9) |
| Urea | 135 mg/dL (NV: 16.6-48.5) |
| INR | 1.5 |
| C-reactive Protein | 68.8 mg/dL (NV: 0.0-0.5) |
| Procalcitonin | 71.2 ng/mL (NV: < 0.05) |

NV: Normal value.
Source: Clinical laboratory data from the ISSS Medical, Surgical and Oncological Hospital.

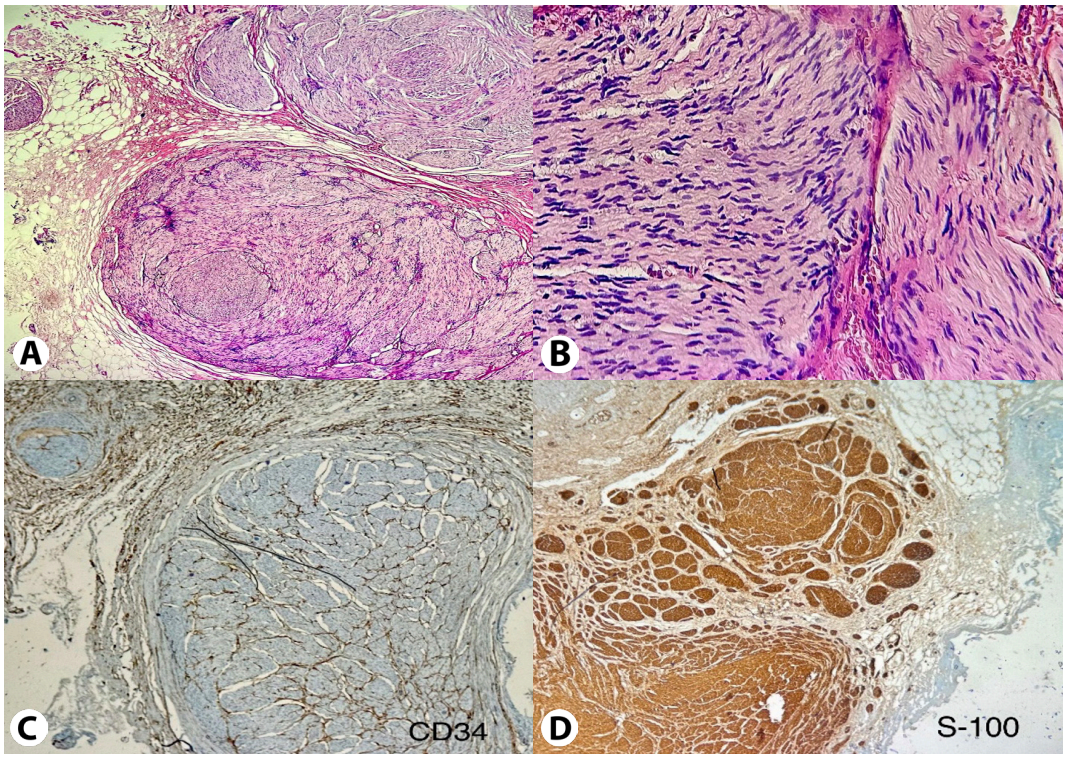


**Figure 4.** A) Panoramic photomicrograph showing a prolifer-ation of cells arranged in a lobular pattern (H&E, 40X). B) Cells with serpentine, wavy nuclei and col-lagen fibrils are observed (H&E, 10X). C) CD 34 demonstrates positive labeling for fibroblastic cells (40X). D) Strong and diffuse positive nuclear and cy-toplasmic expression of S-100 demonstrating prolif-eration of nerve fascicles (40X). Source: Department of Pathology, ISSS

the cases in which patients were diagnosed with traumatic bile duct neuromas occurred after ten years of the surgical event, consistent with other studies.[xi]

Classically, neuromas are divided into two types: primary and traumatic; traumatic neuromas are associated with a surgical event.[vi] This case corresponds to the second type due to the repair process at the site of a traumatic lesion of the peripheral nerves during cholecystectomy, located in the stump of the cystic duct, affecting the main biliary tract.

The diagnosis of a biliary neuroma is challenging due to the nonspecific symptomatology, without typical features.[xii] In addition, efforts are directed to rule out a bile duct cholangiocarcinoma.[xiii] The patient initially presented with acute cholangitis, associated with stenosis of the main bile duct. It was not possible to rule out malignancy by imaging studies; however, the diagnosis was made by histopathological study. The management algorithm is similar to that of a malignant neoplasm, requiring a radical surgical procedure as the treatment of choice. In situations where a malignant lesion is not suspected, the initial approach should be endoscopic or percutaneous intervention, and in cases refractory to treatment and symptomatic, surgery is the indicated procedure.[viii,xiv] Some cases may present with elevated tumor markers (CA19-9), especially in cases of cholangitis; however, it is not useful for differentiating cases suspected of malignancy,[viii] such as the present case, with an elevated value in benign disease.

Histopathological analysis is necessary to make the diagnosis of this rare disease, particularly when there is suspicion of a malignant neoplastic lesion. Macroscopically, the lesions are observed as an ovoid and encapsulated tissue.[xv] In this case, histological analysis showed spindle cells without nuclear pleomorphism and low mitotic activity, and upon immunohistochemistry, they were positive for S100, confirming their neurogenic origin. The immunohistochemistry of the present case was positive for S100 and a Ki67 of 2%. The prognosis of traumatic neuromas is good, with a low recurrence.

## Ethical aspects

This work complies with the standards established in the Declaration of Helsinki and Belmont, respecting patient confidentiality and all ethical aspects. The confidentiality of the information is guaranteed and the informed consent of the patient is obtained for the publication of the information for scientific purposes.


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