RESEARCH ARTICLE

A CASE OF TRAUMATIC NEUROMA OF THE COMMON BILIARY DUCT SIMULATING A CHOLANGIOCARCINOMA

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Abstract

We present a case of traumatic neuroma of the common bile duct in a 53-year-old woman who presented with jaundice, 5 months after cholecystectomy. Physical examination revealed jaundice, epigastric pain and scratching lesions. Gamma glutamyl transferase (GGT) at 109 U/L, Alkaline phosphatase at 175 U/L. CA19-9 was at 500U/ml. Magnetic resonance cholangiopancreatography revealed a T1 and T2 hyperintense lesion of perihilar fat. An increase in intensity was noted after use of gadolinium. Exploratory laparotomy was performed with the presumptive diagnosis of cholangiocarcinoma. Pathologic analysis of the surgical specimen revealed Haphazard mature nerves, consistent with a bile duct neuroma. Follow-up of the patient showed no post-operative complications or signs of recurrence.

Introduction:

Traumatic neuroma is an uncommon cause of bile duct obstruction. It represents reactive proliferation of nerve fibers that are characterized by a neural enmeshment in an overgrowth of the Schwannian sheath cells [1, 3].

Since the bile duct is surrounded by an abundant nerve supply, it can be a location of traumatic neuroma. It occurs after cholecystectomy [2].

The patient usually has a history of surgery to the biliary tract. [4-6]

Traumatic neuromas of the biliary tract are usually asymptomatic, rarely they present with intermittent symptomatic right upper quadrant pain and jaundice.

Cholangiocarcinoma is the leading differential diagnosis. [2]

The best treatment option is surgery, with good long-term results. [6]

Case report:

A 53-year-old woman was admitted to the hospital for the investigation of obstructive jaundice and generalized pruritus.

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History of previous illness includes an open cholecystectomy for cholecystitis 5 months ago. Physical examination revealed jaundice, epigastric pain and scratching lesions. She had no fever and vital signs were within normal limits. White blood cell count was normal. Liver function tests were as following: Total bilirubin at 40.7 mg/dL, direct bilirubin at 28 umol/L, Aspartate transaminase (ASAT) at 86 U/L, Alanine transaminase (ALAT) at 59 U/L; Gamma glutamyl transferase (GGT) at 109 U/L, Alkaline phosphatase at 175U/L, TP at 89 and CRP at 11.55. CA19-9 was at 500U/ml.

Other laboratory values were within the normal range.

Abdominal ultrasonography demonstrated moderately dilated intra and extrahepatic ducts. Magnetic resonance cholangiopancreatography revealed a T1 and T2 hyperintense lesion of peri-hilar fat. An increase in intensity was noted after use of gadolinium.

Intrahepatic biliary ducts were discretely dilated with presence of a short stenosis below the biliary confluence extending over 6 mm. (Figure 1)

Exploratory laparotomy was performed with the presumptive diagnosis of cholangiocarcinoma. At operation many adhesions were found, a resection of extrahepatic bile duct was performed along with a hepatico-duodenal bypass and lymph node dissection of the hepatic pedicle.

Pathologic analysis of the surgical specimen revealed haphazard mature nerves within mature collagenous scar. The observed nerves were made of axons and Schwann cell which lacked atypia or mitotic figures. These findings were consistent with a bile duct neuroma. (Figures 2 and 3)

An immunohistochemical study allowed confirmation of the nervous nature of the observed fibers since they were S-100 positive (Figure 4)

The postoperative course was uneventful, and the patient was discharged 10 days after surgery. Follow-up of the patient showed no post-operative complications or signs of recurrence.

**Discussion:**
Traumatic neuroma of the biliary tree was first described by Husseinoff in 1928.[4]

Cholecystectomy is the most frequently performed operation in abdominal surgery [6]

Neuromas have been most commonly described after both laparoscopic and open cholecystectomy.

They may also occur with common bile duct exploration procedures. [7]

Traumatic nerve cell growth after surgery is thought to be the trigger of Neuroma formation.[4-7]

It is a nonneoplastic disorganized proliferation of axons, Schwann cells, and perineurial cells in a fibrocollagenous stroma.[7]

Sympathetic and parasympathetic fibers arising from the greater and lesser splanchnic nerves are involved in the pathogenesis. [8]

The incidence of traumatic neuroma is unknown since most patients remain asymptomatic.

The biliary tree neuroma presents from several months to 46 years after cholecystectomy [4-8].

Most occur in the cystic duct stump after cholecystectomy and are asymptomatic. Rarely they present with intermittent or continuous symptomatic epigastric pain or right upper quadrant and jaundice. Our patient presented with right upper quadrant pain and jaundice.
Accurate preoperative diagnosis of neuroma can be difficult. Many patients, as in our case, were thought to have an underlying malignancy. [9]

In most cases, the cholangiocarcinoma is the leading differential diagnosis because of the similarity of presentation.[9-14]

Obstructive jaundice is frequently associated with false CA19-9 elevation in benign conditions. Our patient had an elevated CA 19-9 level of 500 U/ml prior to surgery, that is suspected to related to malignancy. [9,10,13]

Radiological explorations show dilated common bile duct and extrahepatic bile ducts. They can very rarely identify the traumatic neuroma as a strongly enhanced lesion, as in our case, on MRImaging or CT-scan[1]

In the majority of cases, a definitive diagnosis is obtained by pathological examination of the surgical resection specimen.

In the literature, a neuroma was recently diagnosed preoperatively by biopsy with cholangioscopy. [8]

Pathologically, the lesion manifests as haphazard mature nerves within mature collagenous scar. The nerves are made of axons and Schwann cell. No atypia or mitotic activity should be observed.

The main treatment is surgical resection of the lesion. No cases of recurrence have been found in the English literature.

**Figures:**

Figure 1. MR Imaging showing a T1 Hyperintense lesion of the fatty tissue around the biliary convergence.
Figure 2. - Microphotography at low power showing a proliferation of nerve fibers (Right side of the picture), compressing the bile duct (Left side of the picture).

Figure 3. - Microphotography showing the bland appearance of the proliferating cells, forming haphazardly organized fibers.
Conclusion:
Awareness of traumatic neuromas should be the rule especially in front of a patient with a history of biliary tree surgery. This may aid preoperative work up and planning, as well as patient counseling.

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., KARIMDIEE B.S., CONVERSESET S., SAINTPAUL 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., KOBYASHI 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. Zeff, R.H., Pfeffer, R.B., Adams, P.X., Ruoff, M., 1976. Reoperation for amputation neuroma of the cystic duct. Am. J. Surg. 131, 369–370.
5. De Rosa, A., Gomez, D., Zaitoun, A.M., Cameron, I.C., 2013. Neurofibroma of the bile duct: a rare cause of obstructive jaundice. Ann. R. Coll. Surg. Engl. 95, e38-40.
6. Larson, D.M., Storsteen, K.A., 1984. Traumatic neuroma of the bile ducts with intrahepatic extension causing obstructive jaundice. Hum. Pathol. 15, 287–289.
7. apovilla, M., Lazure, T., Lorand, I., Carton, E., Rocher, L., Pelletier, G., Cucherousset, J., Bedossa, P., 2005. [Post-cholecystectomy amputation neuroma of the common bile duct with obstructive jaundice]. Gastroenterol. Clin. Biol. 29, 79–81.
8. Cheng, Y., Jia, Q., Xiong, X., Cheng, N., 2014. Traumatic bile duct neuroma after resection of hilar cholangiocarcinoma. Clin. Res. Hepatol. Gastroenterol. 38, 127–128. doi:10.1016/j.clinre.2013.12.007
9. Rastogi A, Bihari C, Arora A, Sinha PK, Chattopadhyay TK, Kishore GS. Hilartraumaticneuromamasquerading as Klatskintumor. Trop Gastroenterol2015;36:54-56.
10. Janes S, Renault PH, Gordon MK. Traumatic (or amputation) neuroma. anz j surg 2004 aug 74(8):701-2
11. Kaman L, Sanyal S, Behera A, Singh R, KatariyaRN. Comparison of major bile duct injuries following laparoscopic cholecystectomy and open cholecystectomy. ANZ J Surg. 2006 Sep;76(9):788-91.
12. Chaudhary A, Manisegran M, Chandra A, Agarwal AK, Sachdev AK. How do bile duct injuries sustained during laparoscopic cholecystectomy differ from those during open cholecystectomy? J Laparoendosc Adv Surg Tech A. 2001 Aug;11(4):187-91.

13. 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.

14. Husseinoff D. Ueber einem Fall von Wucherung des Nervengewebes nach wiederholten Operationen der Gallengänge. Zbl Allg Path 1928;43:344-348.