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CASE REPORT

Contrast-enhanced ultrasound of a traumatic neuroma of the extrahepatic bile duct: A case report and review of literature

Zhi-Qiang Yuan, Hua-Lin Yan, Jia-Wu Li, Yan Luo

Abstract

BACKGROUND

Traumatic neuromas result from nerve injury after trauma or surgery but rarely occur in the bile duct. However, it is challenging to diagnose traumatic neuromas correctly preoperatively. Although some previous reports have described the imaging features of traumatic neuroma in the bile duct, no features of traumatic neuromas in the bile duct have been identified by using contrast-enhanced ultrasound (CEUS) imaging before.

CASE SUMMARY

A 55-year-old male patient presented to our hospital with a 3-mo history of abdominal distension and anorexia and history of cholecystectomy 4 years ago. Grayscale ultrasound demonstrated mild to moderate intrahepatic bile duct dilatation. The lesion approximately 0.8 cm × 0.6 cm with a regular shape and clear margins. The nodule of the bile duct showed slight hyperenhancement in the arterial phase and isoenhancement in the venous phase on CEUS. Laboratory tests showed that alanine aminotransferase and aspartate aminotransferase were increased significantly, while the tumor marker carbohydrate antigen 19-9 was increased slightly. Then, hilar bile duct resection and end-to-end bile ductal anastomosis were performed. The histological examination revealed traumatic neuroma of the extrahepatic bile duct. The patient had an uneventful recovery after surgery.

CONCLUSION

The current report will help enhance the current knowledge regarding identifying traumatic neuromas by CEUS imaging and review the related literature.

Key Words: Traumatic neuroma; Bile duct; Contrast-enhanced ultrasound; Enhancement; Cholangiocarcinoma; Case report
INTRODUCTION

A traumatic neuroma is a chronic reparative proliferative response of the nerve after trauma or surgery. It is composed of disorganized nerve fiber bundles with fibrous stroma, Schwann cells, perineural cells, axons, and endoneurial fibroblasts[1]. The common sites of traumatic neuromas are the necks and extremities[2,3]. Although some studies have described traumatic neuromas in the bile duct, cases of sonographic features of contrast-enhanced ultrasound (CEUS) have not been published before. The clinical manifestation and imaging examination of a traumatic neuroma of the bile duct are not specific, which makes it challenging be accurately diagnosed preoperatively. Herein, we report a traumatic neuroma of the extrahepatic bile duct with detailed ultrasonographic imaging features. We also reviewed the literature on the imaging findings for traumatic neuromas.

CASE PRESENTATION

Chief complaints
A 55-year-old man was admitted to our hospital with unexplained abdominal distension and anorexia 3 mo ago.

History of present illness
The patient suffered from unexplained abdominal distension and anorexia for 3 mo. The patient developed darkened urine 2 mo ago. He experienced a weight loss of 5 kg over the course of the disease. He underwent contrast-enhanced computed tomography (CECT) examination at a local hospital, and a lesion was found in the extrahepatic bile duct, which was believed to be a tumor.

History of past illness
The patient underwent cholecystectomy for gallbladder stones with an uneventful postoperative recovery 4 years ago. He had a 10-year history of hypertension.

Personal and family history
There was no other personal or family history of acute or chronic disease.

Physical examination
The patient showed no tenderness, rebound tenderness or muscle tension on abdominal palpation.

Laboratory examinations
The liver function tests demonstrated increased levels of alanine aminotransferase (185 IU/L, normal range: < 50 IU/L), aspartate aminotransferase (148 IU/L, normal range: < 40 IU/L) and total bilirubin (37.0 μmol/L, normal range: 5 μmol/L to 28 μmol/L). Tumor markers included carbohydrate antigen 19-9 (98.6 U/mL, normal range: < 22 U/mL), carcinoembryonic antigen (0.97 ng/mL, normal range: < 5 ng/mL), and alpha-fetoprotein (4.67 ng/mL, normal range: < 7 ng/mL).

Imaging examinations
The patient underwent an abdominal ultrasound (US) examination by a Resona7 US system (Mindray Medical International, Shenzhen, Guangdong Province, China) equipped with an SC6-1U (1-6 MHz)
transducer. The US revealed mild to moderate dilatation of the intrahepatic bile duct, and the diameter of the upper extrahepatic bile duct was 1.2 cm (Figure 1A). A hyperechoic nodule sized 0.8 cm × 0.6 cm was found in the upper extrahepatic bile duct with an almost regular shape and slightly clear margins (Figure 1B). The patient underwent CEUS with the patient’s consent for further diagnosis. A 2.4-mL US contrast agent SonoVue (Bracco, Milan, Italy) suspension was injected through the left cubital vein followed by a flush with 5 mL saline. In the arterial phase, the nodule showed slight heterogeneous hyperenhancement without rim-like enhancement (Figure 1C). The nodule appeared heterogeneous isoenhancement in the venous phase (Figure 1D). Additional CECT in our hospital showed a hypoenhancement nodule approximately 1.3 cm × 1.0 cm in size in the upper extrahepatic bile duct (Figure 2).

FINAL DIAGNOSIS

Based on the incidence of bile duct diseases, imaging findings and laboratory tests, the patient's clinical diagnosis was hilar cholangiocarcinoma. However, postoperative pathology of the common bile duct lesion showed a neoplastic proliferation of submucosal nerve tissue and fibrous tissue (Figure 3A), and an immunohistochemistry marker was positive for S-100 (Figure 3B). The above pathological findings indicated that the lesion in the bile duct was a traumatic neuroma.

TREATMENT

During the surgery, intraoperative frozen pathology showed no tumor cells within the bile duct lesion. Therefore, hilar bile duct resection and end-to-end bile ductal anastomosis (EE) were performed. The patient recovered uneventfully after surgery.

OUTCOME AND FOLLOW-UP

There was no obvious abnormality on CECT for half a year after the operation.

DISCUSSION

Extrahepatic bile duct masses are commonly malignant tumors, while benign tumors account for only 6% [4-7]. Consequently, the possibility that extrahepatic bile duct lesions are traumatic neuromas is easily overlooked. It has been reported that most traumatic neuromas of the biliary tract arise in the cystic duct stump after cholecystectomy [8]. If a nerve is transected and its continuity cannot be reestablished, a traumatic neuroma may develop [9].

We reviewed the literature from 2000 to 2021 and found 18 publications regarding the imaging features of traumatic neuromas in the bile ducts [2,10-26]. The clinical findings and imaging features of these 18 reported cases are summarized in Table 1. Finally, 22 patients were included in the literature review for further analysis. The age of patients ranged from 17 to 81 years of age, and there was a significant male predominance, with 15 males (68.2%), 2 females, and 5 patients of unreported sex. Most cases were secondary to cholecystectomy, but a few were secondary to liver transplantation, hepatectomy and hilar cholangiocarcinoma. The major symptoms found in these patients were jaundice, abdominal pain, and weight loss, while some patients had no apparent symptoms.

Unfortunately, no specific imaging features for traumatic neuromas of the bile duct have been found at present. Although some imaging modalities, such as US, computed tomography (CT), and nuclear magnetic resonance imaging (MRI), are valuable to some extent, it remains a challenge to diagnose traumatic bile duct neuromas preoperatively [17]. Imaging findings in these 22 patients varied from nodules or masses to localized bile duct stenosis with dilatation of the upper bile duct. It has been reported in the literature that the US imaging findings of extraabdominal nerve tumors and traumatic neuromas are generally hypoechoic masses, larger than the nerve trunk and continuous with the nerve [27]. However, the nerve injury related to cholecystectomy may be too small, so we could not find that the nerve is connected to traumatic neuroma of the bile duct. US was performed in 5 of the 22 patients, 2 of whom showed hypoechoic nodules, and the remaining 3 patients showed stenosis and dilatation of the bile ducts. However, our patient's US sonogram showed a hyperechoic nodule, indicating that the echogenicity of the nodule of traumatic neuroma was variable.

CECT was performed in 2 of the 18 cases, and an enhancing nodule was seen, which was consistent with the CECT findings of our patient. Traumatic neuromas also show enhancement on MRI when a contrast agent is used [28], which may be related to a damaged peripheral nerve blood barrier that occurred during a prior insult to the nerve [29-32]. One of these 18 cases described the enhancement
Figure 1 Ultrasound images of the patient. A and B: The ultrasound (US) showed mild to moderate intrahepatic bile duct dilatation (orange arrow) and a hyperechoic nodule sized 0.8 cm × 0.6 cm (orange arrow) in the extrahepatic bile duct; C and D: In the arterial phase, contrast-enhanced US (CEUS) showed slight hyperenhancement (orange arrow); in the venous phase, CEUS showed isoenhancement (orange arrow).

Figure 2 Contrast-enhanced computed tomography images of the patient. Contrast-enhanced computed tomography showed a hypoenhancement nodule in the upper extrahepatic bile duct (orange arrow).

The diagnosis of bile duct traumatic neuroma was correctly diagnosed in 1 of the 18 cases examined and confirmed by biopsy. The remaining cases were not correctly diagnosed, and it was difficult to distinguish between benign and malignant lesions in most cases. Therefore, surgery would be performed on a large proportion of patients. Once the patient underwent surgery, an intraoperative frozen section examination helped to confirm that the lesion was benign and extensive surgical
### Table 1 Traumatic neuroma of the bile duct reported in the literature between 2000 and 2020

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<th>Ref.</th>
<th>Age</th>
<th>Sex</th>
<th>Symptoms</th>
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<td>Shimura et al[10]</td>
<td>70</td>
<td>F</td>
<td>Abdominal discomfort</td>
<td>Extrahepatic bile duct</td>
<td>US: Hypoechoic tumor, bile duct slightly dilated</td>
<td>Did not indicate bile duct carcinoma</td>
<td>Bile duct excision and a Roux-en-Y hepaticojejunostomy</td>
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<td>CT: Round, hyperdense, distinct margin tumor</td>
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<td>Angiography: No encasement of the surrounding major vessels</td>
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<td>Endoscopic retrograde cholangiography: A protuberant nodule</td>
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<td>Intraductal ultrasonography: A smooth hypoechoic tumor</td>
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<td></td>
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<td>MRCP: A focal stricture</td>
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<tr>
<td>Ueno et al[2]</td>
<td>60</td>
<td>M</td>
<td>Jaundice</td>
<td>Mid-common bile duct</td>
<td>US: Dilatation of the bile ducts, a mildly echogenic mass</td>
<td>Could not confirm benign or malignant nature</td>
<td>Bile duct excision and a hepato-jejunostomy</td>
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<td></td>
<td>CT: Dilatation of the bile ducts, a markedly enhanced nodule</td>
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<td>MRI: Dilatation of the bile ducts. Homogeneous enhanced nodule with an iso-intense to the aorta, both in the arterial and portal phase</td>
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<td>Percutaneous transhepatic cholangiography: Dilatation of the bile ducts and a smooth stricture</td>
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<td>Choi et al[13]</td>
<td>46</td>
<td>M</td>
<td>Increased liver enzymes</td>
<td>Right hepatic duct</td>
<td>CT: A mass approximately 2 cm</td>
<td>A bile duct cancer could not be excluded</td>
<td>Right hemihepatectomy</td>
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<td></td>
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<td>MRI: A mass approximately 2 cm</td>
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<td>Kim et al[14]</td>
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<td>M</td>
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<td>MRC: Eccentric wall thickening of the bile duct consistent with a neoplasm</td>
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<td>M</td>
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<td>Distal</td>
<td>US: Dilatation of bile</td>
<td>Ampullary or</td>
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<tr>
<td>Navez et al [18]</td>
<td>ND</td>
<td>ND</td>
<td>Jaundice (3 patients) or liver function test alteration (1 patient), a retro-obstructive choleperitoneum on the downstream biliary stenosis (1 patient)</td>
<td>MRI: A markedly homogeneous high intensity nodule enhanced on portal-phase (1 patient), anastomotic biliary stricture (4 patients)</td>
<td>ND</td>
<td>Traumatic biliary neuromas resection combined with hepaticojejunostomy (1 patient); traumatic biliary neuromas resection and duct-to-duct biliary reconstruction protected by a T-tube (4 patients)</td>
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<td>Terzi et al [19]</td>
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<td>17</td>
<td>Persistent elevated transaminase and bilirubin levels</td>
<td>Percutaneous transhepatic cholangiography: A biliary stricture at the anastomosis</td>
<td>ND</td>
<td>Resection of the bile duct stricture and a Roux-en-Y hepaticojejunostomy</td>
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<tr>
<td>Toyonaga et al [20]</td>
<td>F</td>
<td>76</td>
<td>A bile duct nodule</td>
<td>CT: An 8 mm, smooth, and uniformly enhanced nodule</td>
<td>Submucosal tumor</td>
<td>Biopsy, observation for 1 year, no changes to the nodule</td>
<td></td>
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<td>Hirohata et al [22]</td>
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<td>60</td>
<td>No chief complaint</td>
<td>US: A 6 mm round tumor, surrounding lymph nodes were not swollen</td>
<td>Cholangiocarcinoma</td>
<td>Surgery</td>
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<td>Yasuda et al [23]</td>
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<td>ND</td>
<td>Stump of the dilated cystic duct</td>
<td>MRI: A slightly high signal on T2 and the periphery remnant cystic duct of the tumor presented as a high-intensity lesion on T2</td>
<td>Amputation neuroma</td>
<td>Biopsy, observation</td>
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Cholangiogram: A hemispherical defect
Cholangioscopy: A hemispherical mass covered with thin normal cystic duct epithelium

Lalchandani et al[24] 41 M Epigastric pain, weight loss, tea-colored urine Common hepatic duct US: Dilation of the bile ducts Acute cholangitis First: Biliary stent Finally: Bile duct resection and hepaticojejunostomy

Kim et al[25] 72 M A duodenal subepithelial tumor during a medical checkup Near the duodenal wall and the cystic duct stump CT: A 1.4 cm mass Duodenal subepithelial tumor Resection of the mass and duodenal wall, en-block resection of the mass and cystic duct origin

Nechi et al [26] 76 M Jaundice The transition zone between the common hepatic duct and the main bile duct US: Dilation of the bile ducts, a 5 mm hypoechoic nodule Could not confirm benign or malignant nature Resection of the main bile duct with a choledocho-duodenal anastomosis

MRI: Dilation of the common hepatic duct

ND: Not described; US: Grayscale ultrasound; CT: Computed tomography; CECT: Contrast-enhanced computed tomography; MRI: Magnetic resonance imaging; MRC: Magnetic resonance cholangiogram; MRCP: Magnetic resonance cholangiopancreatography; EUS: Endoscopic ultrasonography; ERCP: Endoscopic retrograde cholangiopancreatography.

Figure 3 Postoperative histopathological images of the patient. A: Hematoxylin and eosin staining showed proliferation of submucosal nerve tissue (magnification, × 100); B: Immunohistochemical staining displayed S100(+) (magnification, × 100).

resurrection of the traumatic neuroma was avoided[2,3]. The primary treatment reported in the literature consists of bile duct excision and hepaticojejunostomy (HJ). Although HJ is frequently recommended for reconstruction, the indications, surgical options and suture selection are also controversial. Some investigators also recommend EE because it is more physiological and can maintain physiological balance [33]. It is possible to achieve excellent long-term results and high quality of life using both HJ and EE when it is feasible for the proximal and distal ductal ends to permit EE[34]. Therefore, the choice of the optimum method is strictly correlated with the morphological nature of the lesion, which is different from one stage to the other, depending upon the moment of detection, and therefore have different surgical implications[35]. The surgeon found that the anastomosed edges blood supply was good and that there was no tension of the anastomosed edges in this patient. Therefore, according to the actual conditions of patients, as well as to maintain physiological balance, our hospital professor implemented EE for this patient.
In this patient, the symptoms of anorexia, weight loss and jaundice mimicked those often caused by malignant tumors of extrahepatic bile ducts. CEUS and CECT showed enhancement of the nodule. Based on the incidence of bile duct diseases and imaging findings, the surgeons misdiagnosed it as cholangiocarcinoma. Periductal infiltrative cholangiocarcinomas account for the majority of extrahepatic cholangiocarcinomas[36]. Extrahepatic cholangiocarcinomas may show hyperenhancement, isoenhancement, or hypoenhancement in the early phase of CEUS, and most of them show hypoenhancement in the late phase[37]. If we find a nodule in the bile duct, we should rule out the diagnosis of cholangiocarcinoma when the nodule does not show hypoenhancement in the late phase of CEUS. However, when traumatic neuroma presents as localized bile duct stenosis, it is relatively difficult to distinguish it from malignant lesions. When a patient has a history of biliary system surgery and the tumor markers are not significantly elevated, suspicion of traumatic neuroma increases. If conditions permit, patients can be protected from unnecessary surgeries by confirming the diagnosis with a biopsy. CEUS is beneficial for differentiating cholangiocarcinoma from traumatic neuromas, but more cases are needed to summarize the sonographic features of this disease. Recognizing of traumatic neuromas may aid in preoperative work up, planning, and patient counseling[24].

CONCLUSION

It is difficult to correctly diagnose traumatic neuroma of the bile duct before surgery. We should rule out malignant differential diagnoses, such as cholangiocarcinoma preoperatively, to avoid unnecessary surgery. The enhancement mode of CEUS may provide information to distinguish traumatic neuromas from malignant lesions. We need to combine the history of biliary tract surgery, clinical findings, imaging findings and laboratory tests to diagnose this disease.

FOOTNOTES

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imaging is a noninvasive method of observing injury and recovery in the peripheral nervous system.


