Choledocho-nodal Fistula: Uncommon Cause of Obstructive Jaundice in a Patient with HCC Diagnosed by Combined ERCP/EUS

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ABSTRACT
A 58-year-old male patient presented with advanced hepatocellular carcinoma underwent transarterial chemoembolization (TACE) for hepatic focal lesions followed by TACE for a solitary hilar nodal metastasis combined with regorafenib therapy. One month later, the patient developed progressive jaundice. Work-up showed obstructive jaundice with intrahepatic biliary radicles dilatation. The diagnosis and treatment was achieved by combining endoscopic retrograde cholangiopancreatography and endoscopic ultrasonography and showed uncommon cause of obstructive jaundice due to common bile duct compression by a choledocho-nodal fistula following TACE of a metastatic hilar lymph node.

Keywords: Choledochal, Hepatocellular carcinoma, Obstructive jaundice, Transarterial chemoembolization

INTRODUCTION
Obstructive jaundice results from obstruction of the biliary system starting from inside the liver tissue to the duodenal papilla. Causes include luminal obstruction, for example, by stone or cancer, stricture of the wall as in malignancies, and sclerosing cholangitis or compression from outside as in case of malignant lymph nodes (LN) and cancer pancreas.[¹]

In cases of hepatocellular carcinoma (HCC), the development of jaundice may be related to; infiltration of the bile ducts, obstruction of the lumen by dislodged tumor tissues, or compression of bile ducts by metastatic LNs. Furthermore, with progression of HCC and exhaustion of the functioning liver tissue, the jaundice becomes deeper.[¹]

In the current case report, we reported uncommon case that developed obstructive jaundice after performing transarterial chemoembolization (TACE) for a solitary hilar metastatic LN. To the best of our knowledge, this is the 1st time to describe choledocho-nodal fistula as a cause of obstructive jaundice.

CASE REPORT
Here, we present a 58-year-old male patient with multifocal HCC. The patient was treated 3 times with TACE with the last session for a solitary hilar LN that appeared after the first session
of TACE, and afterward, he was kept by his oncologist on regorafenib 40 mg tid. One month before admission, he developed progressive jaundice and was admitted to the hospital for evaluation. On admission, his laboratories showed high serum bilirubin: Total bilirubin 44.4 mg/dl, direct bilirubin 29.9 mg/dl, ALT 49 IU/l, AST 98 IU/l, and serum albumin 3.6 mg/dl while AFP was 12,000 ng/ml. The baseline laboratories and imaging modalities for the patient were lacking because the patient had a follow up file in another center and presented and presented to us with the jaundice episode.

Abdominal US showed dilated common bile duct (CBD), and non-calculous gallbladder distension and a complementary magnetic resonance cholangiopancreatography (MRCP) showed diffusely dilated upper CBD with compression to the mid-lower segment by a cystic lesion probably from the pancreas. Endoscopic retrograde cholangiopancreatography (ERCP) was done, and on filling downstream, the dye besides showing dilated upper CBD and intrahepatic biliary radicles began to collect in a rounded fashion around the strictured middle-lower CBD [Figure 1].

This was not possibly a biliary leak due localized nature of the lesion and a possibility of choledochal cyst was proposed. A plastic stent 12 cm/10 F was inserted with good biliary drainage. However, the dye retained in the lesion.

An upper GIT endoscopy was done after ERCP withdrawal and showed 1st part duodenal ulcer overlying a bulbar swelling suggesting compression. A decision for an endoscopic ultrasonography (EUS) was taken at the same session. EUS showed partially cystic lesion with heterogeneous content around the CBD and surrounding the inserted plastic stent [Figure 2].

Aspiration of the fluid content of that lesion revealed bile that was confirmed also in laboratory analysis, Figure 3]. Fluoroscopy confirmed the reduced lesion size after EUS aspiration [Figure 4].

Follow-up of the patient after stent insertion by bilirubin level showed reduction of the serum total and direct bilirubin to 14.3/10.1 mg/dl after 1 week and to 6.4/4.7 mg after 2 weeks, respectively.

Figure 1: A 58-year-old male patient presented with obstructive jaundice due to choledocho-nodal fistula. Endoscopic retrograde cholangiopancreatography with cholangiogram showing dilated upper common bile duct (CBD) and intrahepatic biliary radicles (short arrow) and a rounded lesion compressing the lower CBD (long arrow) consistent with the necrotic lymph node.

Figure 2: A 58-year-old male patient presented with obstructive jaundice due to choledocho-nodal fistula. Endoscopic ultrasonography image showing partially cystic lesion (long arrow) with heterogeneous content around the common bile duct consistent with the necrotic lymph node, surrounding the inserted plastic stent (short arrow).

Figure 3: A 58-year-old male patient presented with obstructive jaundice due to choledocho-nodal fistula. Endoscopic ultrasonography with aspiration from the necrotic lymph node showing the yellow fluid consistent with bile. The bile source was confirmed on laboratory examination.
The patient was scheduled for follow-up after 3 months for imaging (CT and MRCP), ERCP for removal of the plastic stent, reevaluation, and arrangement for SEMS if needed. Unfortunately, the patient passed out 7 weeks after hospital discharge due to unrelated event.

DISCUSSION

In patients with advanced HCC and Barcelona Clinic Liver Cancer Stage B (multinodular disease), like our patient, palliative TACE is recommended by the current guidelines.[2] The situation of our patient is quite different because it seems that he developed a single large hilar LN and that probably what encouraged the interventional radiologist to plan for nodal ablation with TACE [Figure 5]. Later, the oncologist added the regorafenib, an oral multikinase inhibitor.

Our patient was admitted due to progressive jaundice following nodal injection/regorafenib therapy, and initially, we sought in hepatic decompensation due to regorafenib. After full laboratories and imaging that revealed dilated biliary system, an ERCP was planned. During the procedure, a suspicion on choledochal cyst was rapidly excluded when the dye failed to be drained after limited sphincterotomy stent insertion and this further confirmed when we shifted to EUS which showed a partially cystic lesion with heterogeneous content and the plastic stent was seen within the lesion which is consistent with the injected LN. It is possible that the LN after injection developed necrosis which penetrated and communicated with the CBD. The bile leak inside the necrotic LN made it sizeable enough to compress the CBD and manifest as obstructive jaundice. One other possibility that cannot be ignored is the technical pitfall during TACE. It is also possible that CBD was punctured iatrogenically and communicated with the LN during injection process with the final result of bile leak within. Injuries following TACE procedures particularly biliary injuries are infrequently reported in the literature. Injuries following TACE seem to be related to microcatheter use.[3]

Uncommon causes of obstructive jaundice have been reported in the literature and range from Mirizzi syndrome, intraductal metastases from distant tumors, outside compression by an aberrant hepatic artery, uncommon tumors, and biliary parasites.[4,5] However, to the best of our knowledge, this is the first case of fistulizing necrotic LN communicating and compressing the CBD to be described in the literature.

The case presented here is important from different aspects. First, the clinical point of view. We are not sure that injection of a malignant metastatic LN would favor better outcome in a patient with advanced HCC. In fact, this behavior should be weighted from the risk point of view in comparison to the expected benefits. There is some evidence in the literature that showed survival benefit for combined TACE for both liver lesions and its regional nodal metastases[6] although not supported by any current guidelines. We believe that this patient with advanced HCC was not an ideal candidate for nodal TACE.

Second, the endoscopic technical point of view. It seems that ERCP/EUS use is superior in evaluating CBD pathology than ERCP alone and although this was reported in the literature,[7] it is reemphasized by this case. With ERCP alone, we thought in a biliary leak and choledochal cyst and also good biliary drainage was achieved with insertion of a plastic stent yet with the introduction of EUS, we precisely identified the nature of the lesion and we aspirated its bile content.
Development of ERCP decades back was an era in the management of pancreaticobiliary disorders because it reduced the morbidity, mortality, and cost of surgical interventions. However, the introduction of EUS was a paradigm shift in management of pancreatic disorders. Furthermore, EUS introduction in management of biliary disorders is growing and its use may be adjuvant or a rescue in ERCP failed cases. In our case presented here, EUS completed the hidden part in the diagnosis of this case and we believe that if our unit was not armed with the EUS, we probably missed the diagnosis.

**CONCLUSION**

This is uncommon case of obstructive jaundice due to CBD compression by a choledocho-nodal fistula following injection of a metastatic hilar LN in a patient with HCC. The diagnosis was achieved by combining ERCP and EUS.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

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**How to cite this article** Emara MH, Zaghloul MS, Mahros AM, Ema EH. Choledocho-nodal fistula: Uncommon cause of obstructive jaundice in a patient with HCC diagnosed by combined ERCP/EUS. J Clin Imaging Sci 2021;11:32.