

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: <http://Elsevier.com/locate/radcr>

Case Report

Hepatoduodenal lymph node metastasis mimicking Klatskin tumor in a patient with sigmoid colon mucinous cancer

Hovhannes Vardevanyan PhD^{a,*}, Josef Holzinger MD^b, Rosemarie Forstner PhD^c

^a Department of Diagnostic Imaging, Armenian-American Wellness Center, Sayat-Nova 6, Apartment 11, Yerevan 0001, Armenia

^b Department of Endoscopic Surgery, University Hospital of Salzburg, PMU, Salzburg, Austria

^c Department of Radiology, University Hospital of Salzburg, PMU, Salzburg, Austria

ARTICLE INFO

Article history:

Received 19 December 2016

Received in revised form

19 March 2017

Accepted 11 April 2017

Available online 2 June 2017

Keywords:

Klatskin

Hepatoduodenal

Metastases

Mucinous

Cancer

ABSTRACT

We report a case of a 48-year-old female patient, who presented with abdominal pain, jaundice, and lack of appetite. Ultrasound showed intrahepatic biliary dilatation with retroperitoneal lymphadenopathy. Further magnetic resonance cholangiopancreatography detected Klatskin tumor. Computed tomography (CT) confirmed the Klatskin tumor with liver metastases and retroperitoneal lymphadenopathy. Biopsy from the hepatic lesion identified mucinous adenocarcinoma, likely originating from bile ducts. Endoscopic retrograde cholangiopancreatography was performed 3 times with stents placed in the left and right hepatic bile ducts. Later the patient had hematochezia and was referred to colonoscopy. Tubulovillous adenoma with dysplasia was diagnosed with signs of in situ cancer. Preoperative CT was done for further staging: new pulmonary metastases were discovered. Sigmoid colon was resected. Histopathology verified a poorly differentiated mucinous adenocarcinoma within the tubulovillous adenoma. Intraoperative biopsies of porta hepatis mass resembled metastatic lymph nodes in hepatoduodenal ligament, mimicking Klatskin tumor. Retrospective analysis of CT data demonstrated presence of sigmoid colon tumor.

© 2017 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Klatskin tumor, also known as hilar or central cholangiocarcinoma, is a malignant tumor of the confluence of hepatic ducts. Gerald Klatskin, an American physician, first described this pathology in 1965 [1]. Predisposing factors include primary sclerosing cholangitis with an up to 30-fold increased risk as well as choledochal cysts and parasitic

infections. [2,3]. Klatskin tumors usually present with clinical symptoms such as upper right abdominal pain and weight loss [4]. Jaundice and pale stools usually are signs of significant tumor extent [5]. The vast majority of Klatskin tumors are lethal, and the disease has a very low overall 5-year survival rate, of as low as 1% [6]. The median survival is approximately 6 months [7]. Currently, no effective medical treatment is present, and only radical surgical resection offers the

Competing Interests: The authors have declared that no competing interests exist.

* Corresponding author.

E-mail address: hovovar@gmail.com (H. Vardevanyan).

<http://dx.doi.org/10.1016/j.radcr.2017.04.002>

1930-0433/© 2017 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 1 – Ultrasound image of intrahepatic biliary dilatation with the obstruction in the porta hepatis (green arrow).

possibility for long-term survival [5,8]. Mucinous adenocarcinomas are clinically, morphologically, and molecularly different from other adenocarcinomas of the colon [9]. Mucinous cancer presenting 5%-15% of all colorectal cancers is a histological subtype, in which the tumor cells secrete extracellular mucin, which involves more than 50% of the tumor mass cases [10]. Several studies suggest that mucinous cancer is associated with a poor prognosis [11,12].

Case report

A 48-year-old female patient was admitted with abdominal pain, mild jaundice, and lack of appetite. Laboratory data showed high levels of: gamma-glutamyl transpeptidase, 300 U/L; bilirubin (total), 30.2 mg/dL; alanine aminotransferase, 281 U/L; and alkaline phosphatase, 509 U/L. Ultrasound examination indicated intrahepatic biliary tree dilatation with enlarged retroperitoneal lymph nodes and some stones in the gallbladder (Fig. 1). Further magnetic resonance imaging

including magnetic resonance cholangiopancreatography confirmed the intrahepatic profound bile duct obstruction caused by a mass in the porta hepatis, which was defined as Klatskin tumor (Fig. 2A). MRI also described two liver metastases and retroperitoneal lymphadenopathy (Fig. 2B). Computed tomography (CT) of thorax, abdomen, and pelvis was performed with oral and I/V contrast administration for tumor staging. The tumor of the porta hepatis with intrahepatic biliary dilatation, liver metastases in II/III and IVb segments, and retroperitoneal lymphadenopathy was diagnosed (Fig. 3A). Retrospectively an ill-defined mass which was not mentioned in the CT report at the time of diagnosis was noted that was adjacent to the uterus, possibly a mass within the elongated mobile sigmoid colon (Fig. 3B). Blood tumor markers were also evaluated, which showed elevation of CEA and CA19-9, with 7.4 $\mu\text{g/L}$ and 78 U/mL, respectively. In order to confirm the diagnoses, ultrasound-guided tru-cut biopsy was performed of the largest liver lesion in IVb segment (Fig. 4). Pathology diagnosed middle grade differentiated mucinous adenocarcinoma, more likely originating from bile ducts. Endoscopic retrograde cholangiopancreatography (ERCP) was performed 3 times within a 10-day period, to reduce the jaundice and resolve the biliary dilatation (Figs. 5A and B). Overall, 4 pigtail stents were placed into the right and left hepatic ducts (Fig. 5C). Consequently, neoadjuvant palliative chemotherapy for Klatskin tumor (T2aN1M1—Stage IVb) was performed as a consensus on the tumor board. The patient started the course with 2 cycles of Cisplatin/Gemzar. While receiving chemotherapy, the patient had hematochezia, which indicated the need for colonoscopy. The latter showed a soft bulky mass inside the sigmoid colon, at about 20 cm distally from the anus. Several biopsies were taken, which were identified as tubulovillous adenomas with high-grade dysplasia by pathology. After 1-month follow-up colonoscopy was performed and multiple biopsies were taken from the sigmoid mass. Pathology revealed a tubulovillous adenoma with high-grade epithelial dysplasia and local invasion as pTis. Thus, another staging CT of thorax, abdomen, and pelvis with peroral and I/V contrast enhancement was performed. Multiple small pulmonary metastases

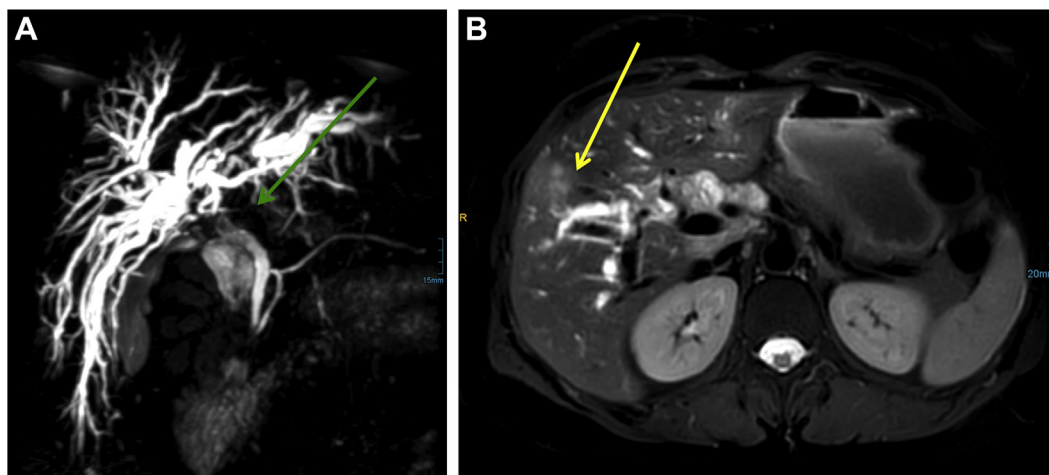


Fig. 2 – Magnetic resonance images of intrahepatic biliary dilatation (green arrow) (A) and irregular hepatic lesions which are hyperintensive on T2-weighted images (yellow arrow) (B).

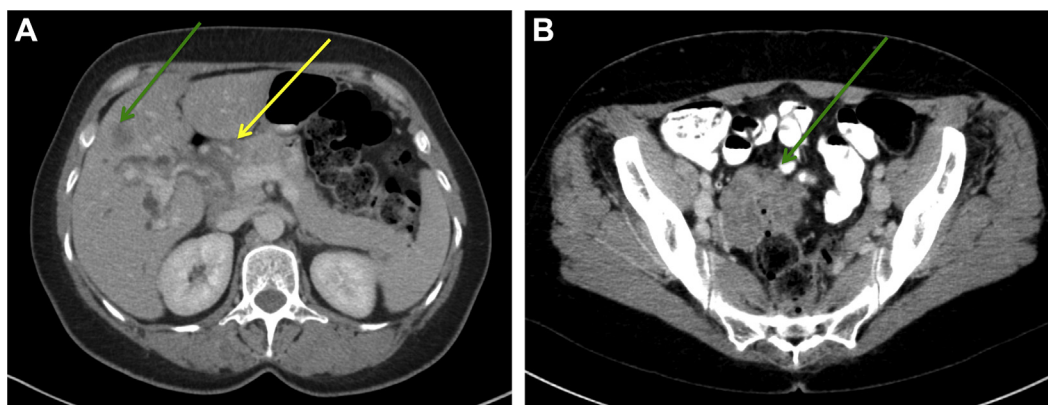


Fig. 3 – CT axial images of a pathologic lesion in the right lobe of the liver (green arrow), with biliary dilatation and a mass in the porta hepatis (yellow arrow) (A). An ill-defined mass is seen in the pelvic cavity on the right (green arrow) (B).

were now identified in both lungs, which were not apparent on the previous CT (Fig. 6). Also, there was enlargement of the tumor in the porta hepatis and of retroperitoneal lymph nodes (Fig. 7A). Metastases of the liver were also slightly enlarging in size, and the biliary hypertension was still present, even though stenting of the common bile duct was performed (Fig. 7B). On retrospective analysis, the mass of sigmoid colon was still present on the pelvic CT images, even though it was still not described in the actual CT report (Fig. 8). Our patient underwent another 2 ERCPs, with additional implantation of bile duct stents. According to the tumor board consensus, the initial diagnosis of Klatskin tumor was assumed to be incorrect, and colon cancer with hepatic metastasis was more reasonable; therefore, the chemoregimen was changed to FOLFOX/Avastin. At this time, CA19-9 has risen to 1140 U/mL. And after 1 cycle, CA19-9 was dramatically lowered to 284 U/mL. Surgical resection of the colon tumor was planned. Pre-surgical CT of thorax, abdomen and pelvis was performed with oral, rectal, and I/V contrast administration. Lung metastases were still present, some cystic degeneration of liver metastases was noted, and bile flow disruption was resolved due to multiple stents and partial regress of the mass in porta hepatis region (Fig. 9A). In addition, an intramural mass in the elongated sigmoid colon was visualized which was about 5 cm

in size. It was easily identified after rectal positive contrast admission (Fig. 9B). Surgical resection of sigmoid colon was performed by end to-end anastomosis. Final histopathologic examination verified a poor differentiated mucinous adenocarcinoma within the tubulovillous adenoma of the sigmoid colon. Intraoperative biopsy was taken from the pathologic mass within the porta hepatis, which showed lymph node metastasis inside the hepatoduodenal ligament, which were mimicking Klatskin tumor by forming a pathologic mass in the region of the hepatic duct confluence and causing severe intrahepatic biliary obstruction.

Discussion

This case presents an interesting clinical constellation, when colon cancer complications manifested first, which were considered to be Klatskin tumor with hepatic metastasis. Colon cancer metastases to hepatoduodenal ligament lymph nodes appear rarely, compared to gastric cancer, that frequently metastasizes to hepatoduodenal lymph nodes and causes mechanical biliary obstruction [13]. Some cases have been described in the literature, where colon cancer has spread to the porta hepatis, causing bile flow obstruction is considered to be a negative prognostic sign. These patients usually die within the next 6 months [14]. In our case, it was mucinous adenocarcinoma of sigmoid colon, which was inadequately diagnosed by histopathology twice because the biopsies were from the dysplastic regions. In a large retrospective analysis of mucinous colorectal cancer cases, it was determined that extrahepatic metastases are more often found, compared with colorectal adenocarcinoma [9]. A rare case of metastatic mucinous adenocarcinoma of the common bile duct was described, when a patient was diagnosed with stimulant cancers of the common bile duct and transverse colon, but histopathology after surgical resection revealed metastatic mucinous cancer in the transverse colon arising from the common bile duct [15]. A similar case of Klatskin tumor metastasis to the colon was described, which was somehow similar to our case because the biliary pathology was evidently diagnosed by radiological imaging, but the colon tumor was not seen before the surgery [6]. Retrospective

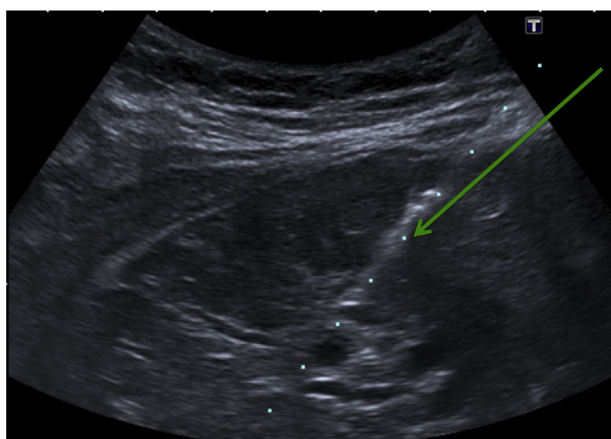


Fig. 4 – Ultrasound image of the biopsy needle passing through liver right lobe lesion (green arrow).

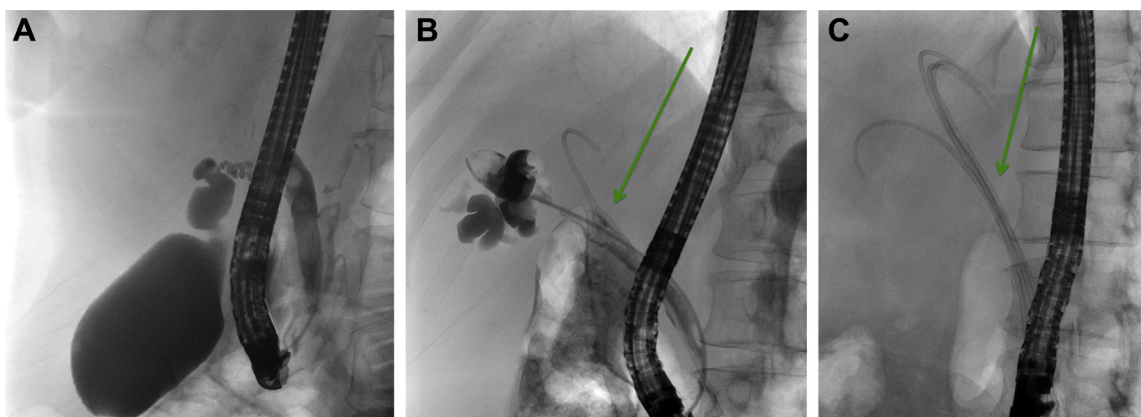


Fig. 5 – Series of ERCP (A), with subsequent insertion of 4 draining stents (C, green arrow). Initially placing two stents (B, green arrow). ERCP, endoscopic retrograde cholangiopancreatography.

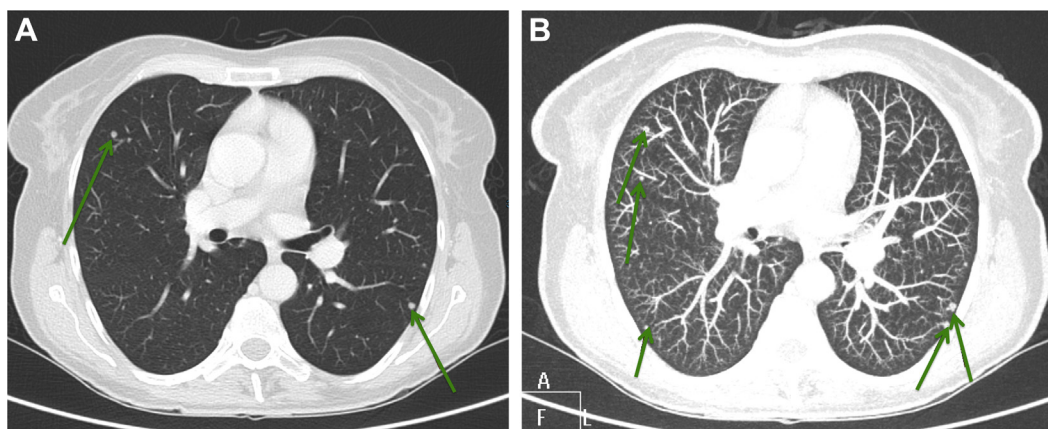


Fig. 6 – Multiple small pulmonary metastasis (A, green arrows), which are easily distinguished on CT MIP-images (B, green arrows).

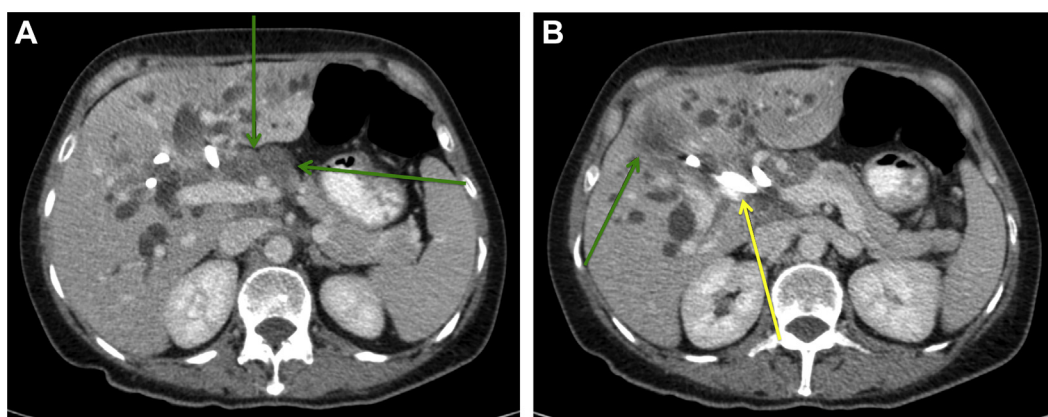


Fig. 7 – CT axial images of porta hepatis tumor (green arrow) (A); liver metastasis (green arrow) and presence of stents in the common bile duct (yellow arrow) (B).

analysis of our patient's radiological data was done. It was noticeable that on first 2 CT examinations, an ill-defined mass in the pelvic cavity was apparent, which was adjacent to the uterus but was not described. Another factor in abdominal

CT imaging is the pre-examination dominance of the clinical manifestation, that is, the biliary obstruction, which was seen previously by magnetic resonance cholangiopancreatography, leads the examining specialist to be

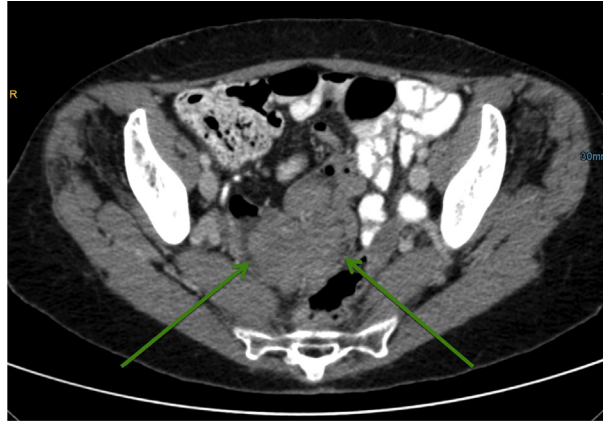


Fig. 8 – A soft tissue mass with irregular borders (green arrows) is visualized in the pelvic cavity, which is adjacent to the uterus.

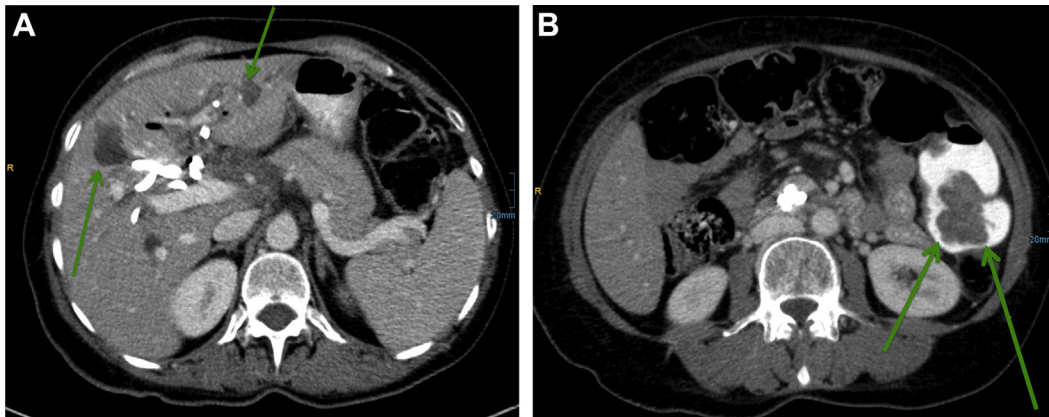


Fig. 9 – CT axial image of liver metastasis with signs of cystic degeneration (A, green arrows). Intraluminal mass of distal sigmoid colon (B, green arrows).

more focused on the biliopancreatoduodenal region. While our clinical case can be recognized as a distinctive case of massive hepatoduodenal lymphadenopathy originating from the sigmoid colon cancer, which clinically presented with jaundice and with simultaneous hepatic metastasis, we suggest using routine rectal contrast administration in addition with oral and I/V contrast enhancement in all oncological cases of abdominal CT imaging, so the radiologist will have more accurate visualization of the bowel lumen.

REFERENCES

- [1] Klatskin G. Adenocarcinoma of the hepatic duct at its bifurcation within the porta hepatis. An unusual tumor with distinctive clinical and pathologic features. *Am J Med* 1965;38:241–56.
- [2] Nagorney DM, McPherson GA. Carcinoma of the gallbladder and extrahepatic bile ducts. *Semin Oncol* 1988;15:106–15.
- [3] Rosen CB, Nagorney DM, Wiesner RH, Coffey Jr RJ, LaRusso NF. Cholangiocarcinoma complicating primary sclerosing cholangitis. *Ann Surg* 1991;213:21–5.
- [4] Schmeding M, Neumann U, Neuhaus P. Colonic metastasis of Klatskin tumor: case report and discussion of the current literature. *World J Gastroenterol* 2006;12(33):5393–5.
- [5] Friman S. Cholangiocarcinoma—current treatment options. *Scand J Surg* 2011;100:30–4.
- [6] Han JK, Choi BI, Kim TK, Kim SW, Han MC, Yeon KM. Hilar cholangiocarcinoma: thin-section spiral CT findings with cholangiographic correlation. *Radiographics* 1997;17:1475–85.
- [7] Park J, Kim MH, Kim KP, do Park H, Moon SH, Song TJ, et al. Natural history and prognostic factors of advanced cholangiocarcinoma without surgery, chemotherapy, or radiotherapy: a large-scale observational study. *Gut Liver* 2009;3:298–305.
- [8] Witzigmann H, Wiedmann M, Wittekind C, Mössner J, Hauss J. Therapeutical concepts and results for Klatskin tumors. *Deutsches Arzteblatt International* 2008;105:156–61.
- [9] Mekenkamp LJ, Heesterbeek KJ, Koopman M, Tol J, Teerenstra S, Venderbosch S, et al. Mucinous adenocarcinomas: poor prognosis in metastatic colorectal cancer. *Eur J Cancer* 2012;48:501–9.
- [10] Symonds DA, Vickery AL. Mucinous carcinoma of the colon and rectum. *Cancer* 1976;37:1891–900.
- [11] Negri FV, Wotherspoon A, Cunningham D, Norman AR, Chong G, Ross PJ. Mucinous histology predicts for reduced fluorouracil responsiveness and survival in advanced colorectal cancer. *Ann Oncol* 2005;16:1305–10.

-
- [12] Catalano V, Loupakis F, Graziano F, Torresi U, Bisonni R, Mari D, et al. Mucinous histology predicts for poor response rate and overall survival of patients with colorectal cancer and treated with first-line oxaliplatin- and/or irinotecan-based chemotherapy. *Br J Cancer* 2009;100:881–7.
- [13] Moon SG, Han JK, Kim TK, Kim AY, Kim TJ, Choi BI. Biliary obstruction in metastatic disease: thin-section helical CT findings. *Abdom Imaging* 2003;28:45–52.
- [14] Thomas JH, Pierce GE, Karlin C, Hermreck AS, MacArthur RI. Extrahepatic biliary obstruction secondary to metastatic cancer. *Am J Surg* 1981;142:770–3.
- [15] Lee DH, Ahn YJ, Shin R, Lee HW. Metastatic mucinous adenocarcinoma of the distal common bile duct, from transverse colon cancer presenting as obstructive jaundice. *Korean J Hepatobiliary Pancreat Surg* 2015;19(3):125–8.