Recurrent biliary hemorrhage caused by metastasis of a malignant small round cell tumor in the common bile duct: a case report

MSRCT in the common bile duct

Ying-Li Jin, Ye-Jiao Ruan, Guang-Rong Lu

Abstract

BACKGROUND
Malignant small round cell tumor (MSRCT) metastasis to the common bile duct and associated with recurrent biliary hemorrhage is extremely rare. Thus far, there have been no reports of metastatic small round cell tumors of the common bile duct in the past, which is the first case.

CASE SUMMARY
Herein, we report the case of a 77-year-old female patient with an MSRCT in the common bile duct. The patient was admitted to hospital due to gastrointestinal hemorrhage and abdominal pain. We found a neoplasm in the common bile duct with active bleeding through a spyglass. We performed biopsy through the spyglass and placed a metal stent to stop bleeding. The pathological result suggested that it was an MSRCT transferred from the back to the common bile duct. Later, we found using fluorescence in situ hybridization that the SS18 gene break test was negative, ruling out the diagnosis of synovial sarcoma.

CONCLUSION
MSRCT is a group of tumors with similar cell morphology and diffuse histological structure. Complete tumor resection results in improved survival in patients with MSRCT. Roux-en-Y cholangiojejunoanostomy was performed. After excision of the common bile duct tumor, the patient felt that the abdominal pain improved and hemorrhage disappeared. The patient underwent routine fecal examination one month after surgery, indicating a negative fecal occult blood test. On May 22nd, 2023, the patient was reexamined with abdominal CT, no abdominal space occupying lesions and abdominal lymphadenopathy were found.

**Key Words:** Common bile duct metastasis tumor; hemorrhage; Recurrent biliary hemorrhage; Malignant small round cell tumor; SpyGlass

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**Core Tip:** Malignant small round cell tumors (MSRCTs) in the common bile duct has not been previously documented. This study aimed to present the case of an MSRCT in the common bile duct. A 77-year-old female patient was admitted to hospital due to gastrointestinal hemorrhage and abdominal pain. We found a neoplasm in the common bile duct with active bleeding through a spyglass. We performed biopsy through the spyglass and placed a metal stent to stop bleeding. The pathological result suggested that it was an MSRCT transferred from the back to the common bile duct.

**INTRODUCTION**

A 77-year-old female patient was admitted to the hospital because of a 1-month history of gastrointestinal hemorrhage and abdominal pain. The patient had severe abdominal pain, no radiation-induced pain, and no nausea or vomiting. The patient defecated hemorrhage once a day. The symptoms of abdominal pain were slightly
eased approximately 20 min after defection. The laboratory examinations showed that the patient's hemoglobin level was decreased to 58 g/L. We performed endoscopic retrograde cholangiopancreatography (ERCP) on the patient. The gastroscope showed that the gastroduodenal mucosa was smooth and pale. The main nipple was found to be changed after EST without blood stains. The nipple was incised with a knife, and dark red liquid was discharged (Figure 1A). On duodenoscopy, the scope was successfully passed through the esophagus, stomach and duodenal bulb, and reached the descending part, and the main nipple was found and was intubated successfully. The amount of contrast agent used was 20 mL. X-ray showed that the common bile duct had developed without expansion, and the maximum diameter was approximately 0.8 cm. It was filled with filling defects of different sizes. The intrahepatic bile duct was partially developed without expansion. There were irregular filling defects in the middle and lower segments of the common bile duct, which could be moved. The bright red liquid was discharged. The cystic pancreatic duct was not developed. The biliary tract was cleared through the zebra guide wire airy catheter, and the bleeding clots and necrotic tissues were removed. After insertion of the spyglass and after repeated flushing, new organisms could be seen in the middle of the common bile duct, with a rough surface, necrosis of the surface, active bleeding and local lumen stenosis (Figure 2C). Spy-Bite was performed. Then, the storage guide wire was inserted into the biliary full posterior membrane metal stent (Figure 1B). After release, the stent was in a good position. Subsequently, the pathological result suggested that it was an MSRCT. Roux-en-Y cholangiojejunostomy and loosening of intestinal conglutination were performed. The specific operation was as follows: first, the abdominal cavity was opened, the middle and upper segment of the common bile duct were expanded, with a diameter of approximately 1.3 cm, there was a palpable mass in the common bile duct approximately 2 cm below the confluence of the left and right hepatic ducts, there was a medium texture and no enlarged lymph nodes were found around it. Then, the bile duct was cut, and a circular yellow and white cauliflower-like new organism was found in the common bile duct, with approximately 2/3 circles around the
common bile duct. The bile duct lesions were removed. On the premise of clear distal patency of the common bile duct, the common bile duct was transected to prepare for the establishment of a new biliary intestinal channel. The distal end of the common bile duct was closed, and the proximal end of the common bile duct was temporarily clamped with nondestructive forceps to prevent bile from flowing into the abdominal cavity. Then, the upper segment of the jejunum was cut off, the transverse colon was lifted down along its mesangium, and the duodenal jejunal flexure was found. The first jejunal artery was kept on the jejunal mesentery, the second jejunal artery was cut off, the jejunal mesentery was separated, cut and ligated so that the distal jejunum had enough freedom, and there was no tension after the abovementioned choledochojejunostomy. The free distal jejunum was sutured and closed and then lifted to the porta hepatitis through the colon for anastomosis. The distal jejunum was lifted 60 cm and anastomosed with the proximal jejunum side to side. A small opening on the side of the opposite edge of the mesentery with the sutured stump was made into the distal jejunum lifted from the transverse mesocolon fissure. The direction was parallel to the long axis of the intestinal tube, the size corresponded to the repaired bile duct orifice and the incision was anastomosed with the jejunum. Finally, the abdominal cavity was closed after the drainage tube was placed. The pathological result suggested that it was an MSRCT with some cytoplasm and difficult-to-see karyokinesis (Figure 2D). Tumors of epithelial cell and neuroendocrine origin were excluded by immunohistochemistry. The pathological examination of bile duct space-occupying lesions showed that they were positive for desmin, vimentin, and CD68 and negative for myogenin, CAM5.2, and S-100. This patient underwent resection of MSRCT in the left shoulder, back and left supraclavicle 9 years previously. No chemotherapy was taken after the operation. Recently, the tumors in the left shoulder, back and left supraclavicle metastasized to the common bile duct. The pathological result suggested MSRCT. The cell morphology and immunohistochemical results were basically consistent with the pathological results of the left shoulder, back and left supraclavicle tumors in 2014. The patient was diagnosed with metastasis of the MSRCT
of the common bile duct. Subsequently, we performed fluorescence in situ hybridization (FISH) and found that the SS18 gene break test was negative (Figure 2E). Therefore, the diagnosis of synovial sarcoma was excluded. After excision of the common bile duct tumor, the patient felt that the abdominal pain had improved, and the hemorrhage had disappeared. The patient underwent routine fecal examination one month after surgery, and the fecal occult blood test was negative. On May 22nd, 2023, the patient was reexamined with abdominal CT, and no abdominal space-occupying lesions or abdominal lymphadenopathy were found.

1 CASE PRESENTATION

Chief complaints
A 77-year-old Chinese female presented to the gastroenterology clinic with a complaint of gastrointestinal hemorrhage and abdominal pain for 1 month.

History of present illness
Symptoms started 1 mo before presentation with gastrointestinal hemorrhage and abdominal pain. The patient felt severe abdominal pain, no radiation-induced pain, and no nausea and vomiting. The patient defecated hemorrhage once a day. The symptoms of abdominal pain were slightly eased about 20 min after defecation.

History of past illness
Nine years ago, this patient underwent the resection of MSRCT in the left shoulder, back and left supraclavicle. No chemotherapy was taken after operation. Recently, the tumors in the left shoulder, back and left supraclavicle metastasized to the common bile duct.

Personal and family history
The patient denied any family history of malignant tumours.
**Physical examination**

On physical examination, the vital signs were as follows: Body temperature, 36.6°C; blood pressure, 97/59 mmHg; heart rate, 84 beats per min; respiratory rate, 16 breaths per min. The abdomen is flat, without tenderness or rebound pain.

**Laboratory examinations**

The laboratory examinations showed that the patient's hemoglobin level decreased to 58 g/L. Fecal occult blood test was positive. Levels of serum tumour markers were normal. No abnormalities in liver and kidney function.

**Imaging examinations**

Abdominal CT and gastrointestinal endoscopy showed no abnormalities.

**FINAL DIAGNOSIS**

Combined with the medical history, the patient was diagnosed with metastasis of the MSRCT of the common bile duct. Later, we found using fluorescence in situ hybridization that the SS18 gene break test was negative, ruling out the diagnosis of synovial sarcoma.

**TREATMENT**

Roux-en-Y cholangiojejunostomy and loosening of intestinal conglutination were performed. The specific operation is as follows: first, open the abdominal cavity, expansion of the middle and upper segment of the common bile duct, with a diameter of about 1.3cm, palpable mass in the common bile duct about 2cm below the confluence of the left and right hepatic ducts, medium texture and no enlarged lymph nodes are found around it. Then cut the bile duct, a circular yellow and white cauliflower like new organism is found in the common bile duct, about 2/3 circles around the common bile duct. Remove the bile duct lesions. On the premise of clear distal patency of the common bile duct, the common bile duct was transected to prepare for the
establishment of a new biliary intestinal channel. Close the distal end of the common bile duct, and temporarily clamp the proximal end of the common bile duct with non-destructive forceps to prevent bile from flowing into the abdominal cavity. Then, cut off the upper segment of jejunum, lift the transverse colon down along its mesangium, and find the duodenal jejunal flexure. Keep the first jejunal artery on the jejunal mesentery, cut off the second jejunal artery, separate, cut and ligate the jejunal mesentery, so that the distal jejunum has enough freedom, and there is no tension after the above-mentioned choledochojejunostomy. The free 3 distal jejunum was sutured and closed, and then lifted to the porta hepatis through the colon for anastomosis. The distal jejunum was lifted 60 cm and anastomosed with the proximal jejunum side to side. The distal jejunum lifted from the transverse mesocolon fissure was cut a small opening on the side of the opposite edge of the mesentery with the sutured stump. The direction was parallel to the long axis of the intestinal tube, and the size was corresponding to the repaired bile duct orifice, and was anastomosed with it. Finally, the abdominal cavity was closed after the drainage tube was placed.

OUTCOME AND FOLLOW-UP

After excision of the common bile duct tumor, the patient felt that the abdominal pain improved and hemorrhage disappeared. The patient underwent routine fecal examination one month after surgery, indicating a negative fecal occult blood test. On May 22nd, 2023, the patient was reexamined with abdominal CT, no abdominal space occupying lesions and abdominal lymphadenopathy were found.

DISCUSSION

MSRCT is an extremely rare malignant tumor that has been clearly defined over the last 10 years. MSRCT is a group of tumors with similar cell morphology and a diffuse histological structure. Complete tumor resection results in improved survival in patients with MSRCT. Chemotherapy and radiotherapy correlate with improved patient outcomes. Multimodal therapy may improve survival in patients with
MSRCTs include Ewing’s sarcoma, peripheral neuroectodermal tumor, rhabdomyosarcoma, synovial sarcoma, non-Hodgkin’s lymphoma, retinoblastoma, neuroblastoma, hepatoblastoma, and nephroblastoma or Wilms’ tumor. Regarding the thoracic cavity, cases involving the pleural, heart, colon, or lungs have been described. To date, no relevant study has reported MSRCTs of the biliary tract. This clinical case report is the first report worldwide of MSRCT metastasized from the left shoulder, back and left supraclavicle to the common bile duct. In this case report, we used a SpyGlass to detect a neoplasm in the patient’s common bile duct and obtained the tissue for pathological diagnosis. The findings showed that a spyglass had an irreplaceable important role in the diagnosis of bleeding in the bile duct. Furthermore, the general morphology of MSRCT in the common bile duct was visually displayed using the spyglass. For biliary bleeding, in addition to considering common causes (such as trauma and biliary infection), we should also consider whether biliary tumors are present. We need to carefully check for the possibility of MSRCT metastasizing to the biliary tract, especially for patients with a history of MSRCT in the past. This study recognized an extremely rare metastasis of MSRCT in the common bile duct and provided new insight into the difficult diagnosis of biliary bleeding in clinical practice in the future.

CONCLUSION
Malignant small round cell tumors (MSRCTs) in the common bile duct has not been previously documented. This study aimed to present the case of an MSRCT in the common bile duct. A 77-year-old female patient was admitted to hospital due to gastrointestinal hemorrhage and abdominal pain. We found a neoplasm in the common bile duct with active bleeding through a spyglass. We performed biopsy through the spyglass and placed a metal stent to stop bleeding. The pathological result suggested that it was an MSRCT transferred from the back to the common bile duct. Although MSRCT is clinically rare, doctors should make a positive and accurate diagnosis and provide humane care and treatment because patients have experienced both mental and
physical problems. We hope that this case report can be accepted by your journal and provide new ideas for clinical work. Let more clinical workers know more about this kind of tumor.
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