

Spontaneous Cholecystocutaneous Fistula in a Case of Adenocarcinoma of Gall Bladder

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Biliary fistulas occur mostly as a result of postoperative complications of liver and biliary tract surgery or trauma¹. Spontaneous external biliary fistulas can rarely occur due to intrahepatic abscess, necrosis or perforation of the gallbladder, or other inflammatory process involving the biliary tract.

Case Report

A 56-year-old female presented with complaints of recurrent episodes of pain abdomen for nine years. The pain was mild to moderate in intensity in the right hypochondrium. The patient was evaluated with a USG abdomen and was diagnosed to have gall stones. The patient denied surgical treatment and was taking over the counter analgesics for pain. The patient had an increase in severity and frequency of pain episodes for two years requiring iv analgesics and hospitalization. The pain was associated with episodes of fever and vomiting. One year ago, she developed a swelling over the anterior abdominal wall in the right upper quadrant associated with pain and fever.

She was evaluated with a CECT abdomen at a peripheral hospital and was being managed with antibiotics

and analgesics. The swelling gradually increased in size, along with the development of erythema. Two months later, the patient had spontaneous rupture of swelling. There was purulent discharge from the wound, which persisted for two months, followed by the spontaneous passage of stone of size around 1x1.5 cm. The fistula healed after 15 days of passage of the stone.

The patient presented to our hospital with a healed fistula scar in the right upper quadrant of the abdomen because of persistent pain abdomen. On general examination, the patient had palpable left supraclavicular lymph nodes. Abdominal examination revealed a palpable gall bladder lump. She underwent an USG abdomen, which showed a contracted gall bladder with irregular wall thickening. On CECT abdomen asymmetric GB wall thickening with infiltration of adjacent liver (seg 4b and 5) and infiltration of abdominal wall was seen. (**Figure 1**)

She was further evaluated with a whole-body PET scan, which revealed tracer uptake in the GB wall with infiltration of the anterior abdominal wall with uptake in bilateral supraclavicular nodes (SUV Max- 13.4). There was no metabolic activity elsewhere in the body. Because of clinically palpable supraclavicular nodes along with uptake on PET scan, USG guided FNAC was done from left supraclavicular node which was suggestive of tubercular lymphadenitis. Ultrasoundguided FNAC was also done from GB wall thickening, which was suggestive of adenocarcinoma.

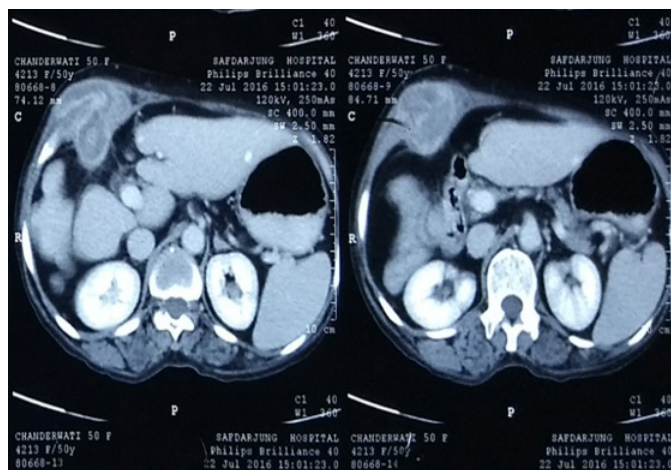


Figure 1: CECT Abdomen showing SCF with abdominal wall collection.

She was started on antitubercular treatment (ATT), one month following which neoadjuvant chemotherapy (NACT) was started. She received four cycles of Cisplatin and Gemcitabine based chemotherapy. Repeat whole-body PET scan done three weeks after completion of chemotherapy, showed a decrease in metabolic activity in cervical lymph nodes (SUV max – 7.8) with a decrease in metabolic activity in GB wall thickening. There were no FDG avid lesions elsewhere to suggest metastases. After pre-anaesthetic evaluation and optimization, patient was planned for definitive surgical resection.

Intra op findings

She underwent staging laparoscopy (**Figure 2**), which showed locally advanced gall bladder cancer and no distant metastasis. On laparotomy gall bladder was contracted with thickening in the fundus and body of gall bladder with infiltration of the anterior abdominal wall, including the right costal margin and diaphragm. She underwent radical cholecystectomy with standard lymphadenectomy with excision of parietal wall containing part of right lower costal margin and diaphragm, including the fistula tract. Diaphragmatic defect was repaired primarily, and onlay prolene mesh closure was done for the abdominal wall defect. The intraoperative frozen section for the cystic duct margin was negative for malignancy.

Histopathology

Histopathology of resected specimen showed mucin secreting adenocarcinoma arising from fundus and body of gall bladder. The tumour was infiltrating into skeletal muscle (anterior abdominal wall) and diaphragm. The healed fistula tract did not show any evidence of tumour cells (**Figure 3**). The cystic duct margin was negative for malignancy. Nine lymph nodes isolated were negative for malignancy. There was no lymphovascular or perineural invasion.

Follow up

The patient was started on adjuvant chemotherapy. CECT abdomen after four months of surgery did not show any evidence of local recurrence or metastatic disease.



Figure 2: Diagnostic laparoscopy.

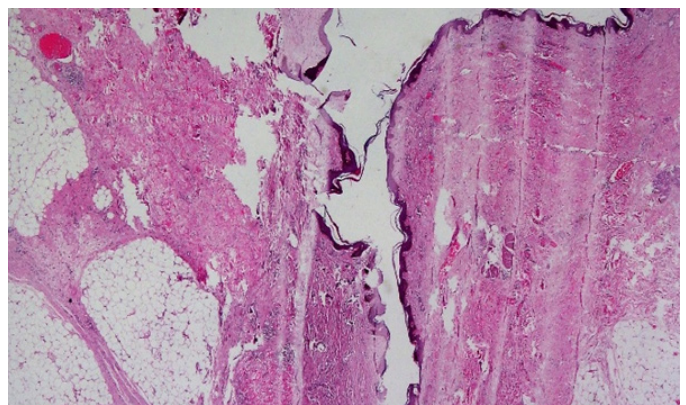


Figure 3: Fistulous tract free of tumor.

Discussion

SCF result from chronic untreated inflammatory pathology secondary to gall stones, although fistulae are reported in the absence of gall stones.^{1,2}

Stone impacted in the cystic duct causes obstruction, which results in distention and increased pressure within the gall bladder. With an increase in pressure, there is impaired blood flow in the gall bladder wall, especially in the fundus, which is most distant from the cystic artery. This may lead to focal necrosis with perforation and eventual abscess formation. When this process continues in untreated cases, the abscess may rupture into the surrounding structures forming internal fistulae with duodenum, colon, stomach, or externally into the abdominal wall, of which later is uncommon. Parietal wall abscess may rupture to the exterior resulting in a cholecystocutaneous fistula.^{3,4}

GB malignancy accounts for 80-90% of all biliary tract cancers. It is an aggressive malignancy with poor overall survival. An SCF, in the case of carcinoma gall bladder, is exceedingly rare with only 3 cases of SCF in patients with GB cancer are reported to date.

In addition to GB wall thickening on imaging, which was suspicious of malignancy, the patient had palpable supraclavicular nodes that were metabolically active on a whole-body PET scan without any metabolic activity in regional abdominal lymph nodes. In this case, metastatic involvement of supraclavicular nodes will make the tumour unresectable. Given suspicion for metastasis, USG guided supraclavicular lymph node FNAC was done. However, when FNAC was done, metabolic activity seen on PET scan in the supraclavicular was indeed due to tubercular lymphadenitis with the presence of granulomas. We previously described the association of tuberculosis in cases of GB cancer, which can influence staging and further management. Seven patients had tuberculosis in association with carcinoma gall bladder. Two patients had supraclavicular lymph nodes, two patients were detected to have TB in inter aortocaval nodes, and one had peritoneal tubercular nodules. Two patients had tuberculosis in dissected hepatoduodenal ligament lymph nodes. At least five of the seven patients would have been deemed unresectable if sampling of nodes was not done. Hence, FNAC and lymph nodes sampling need to be done when IAC or left supraclavicular nodes are involved in a case of carcinoma gallbladder in an otherwise resectable disease.⁵

The patient had a decrease in GB mass metabolic activity, after which the patient underwent definitive surgical resection after initiating ATT and starting NACT. This is the only reported case of SCF associated with adenocarcinoma GB, which has been treated with definitive surgical resection. It has been eight months since the diagnosis of malignancy has been established, and the patient has completed four months of postoperative follow-up and is recurrence-free.

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Nasobiliary Drainage for Intrahepatic Cholestasis and Pruritus Refractory to Medical Therapy: A Series of Three Cases

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Benign recurrent intrahepatic cholestasis (BRIC) and acute viral hepatitis (AVH) are two examples of conditions that can result in intrahepatic cholestasis. Antihistaminics and cholestyramine are generally used for managing pruritus. Some cases have severe pruritus refractory to