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Case Report

Consequences of gallbladder inflammation: spontaneous cholecystocutaneous fistula: a case report

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ABSTRACT

One of the most unusual complications in gall bladder disease is spontaneous cholecystocutaneous fistula (SCF), which has only been reported a few times in the literature. We report the case of a 67-year-old man who presented with a right hypochondrium discharging sinus. Identification of a cholecystocutaneous fistula was made by computed tomography with contrast media, followed by MRCP. This confirmed the presence of a fistulous pathway between the gallbladder and the skin. The patient underwent exploratory laparotomy with subtotal cholecystectomy with en block aponeurotic muscle, skin and fistula orifice excision.

Keywords: Cholelithiasis, Gallbladder inflammation, Spontaneous cholecystocutaneous fistula

INTRODUCTION

Spontaneous cholecystocutaneous fistula (SCF) is an exceptionally uncommon complication of gall bladder disease.

Thilesus first described the condition in 1670 and since 2007 there have only been 28 cases published.[1]

Due to availability of increasingly sophisticated medical imagining techniques and higher standards of public health in general, the incidence of such cases is expected to have markedly declined.

Wider acceptance of laparoscopic cholecystectomy has brought about a revolution in management of the cholelithiasis. Nonetheless, occasionally in neglected, compromised, elderly, cholelithiasis patients with long standing disease, such rare complication can occur.

We report a case of 67-year-old patient who presented with a discharging sinus in right upper quadrant and SCF.

CASE REPORT

Initial presentation and history

We present a case of 67-year-old man presenting with chief complaints of pain and swelling over the right upper abdomen for 30 days, and discharge from right upper abdomen for 25 days. There was no history of fever. The patient reported having the cholelithiasis for 10 years. He had episodes of abdomen pain 4 years earlier which subsided after taking medication. The patient had past history of head injury, craniotomy and convulsions 20 years earlier and he was on antiepileptics and antihypertensive. He also had ischemic heart disease and had previously undergone percutaneous transluminal coronary angioplasty performed in 2012.

Physical examination

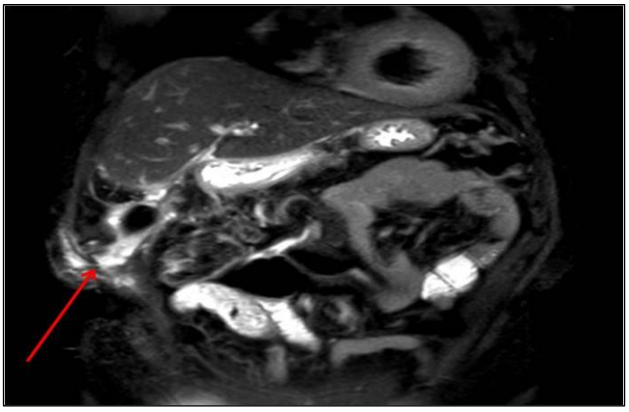
The patient was severely hypertensive (blood pressure 180/100 mmHg) and presented with pallor. On per abdomen examination approximately 10 cm × 8 cm soft

to firm mass in right hypochondrium, mildly tender, and sinus opening with serous like discharge. Surrounding skin was indurated but non-tender and local temperature was not raised.

Laboratory examination and radiological study

Blood test revealed haemoglobin 7 mg/dl, white blood cell count was 18,000/microliter, C-reactive protein was 80 mg/L, international normalized ratio was 4, liver function test unremarkable except raised alkaline phosphatase. Contrast Enhanced Computed Tomography (CECT) revealed a thick-walled gall bladder and thick walled enhancing fistulous tract extending from gall bladder to anterior upper abdominal wall.

Magnetic resonance cholangiopancreatography (MRCP) revealed a track running from base of the gall bladder to anterior upper abdominal wall and not communicating with common bile duct, duodenum, colon, stomach and confirmed with gadolinium sinogram. A single calculus was noted as a well-defined defect in gall bladder (Figure 1 and 2). Also, 2D-Echocardiography showed 45% ejection fraction with Grade 3 left ventricle dysfunction.



The red arrows show the hyperintense cholecystocutaneous fistulous tract.

Figure 1- MR T2W coronal image.

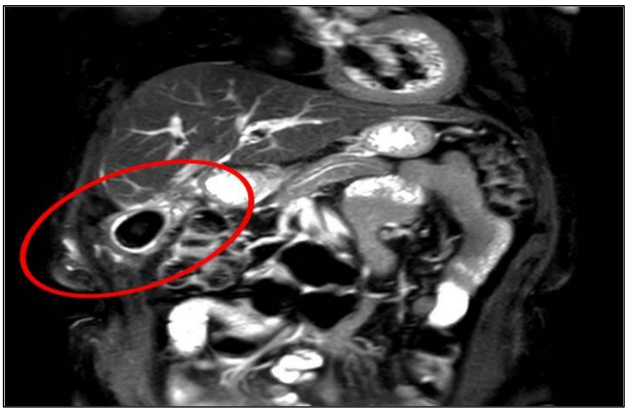


Figure 2: MR T2W coronal image: Hypointense large gall stone.

Treatment and outcome

Patient was started on broad spectrum intravenous antibiotics and anti-hypertensive. Fresh-frozen plasma transfusion transfusion done for deranged INR. Blood transfusion done for anaemia. After initial stabilization patient planned for surgery on the second day after diagnosis. The patient underwent exploratory laparotomy performed through subcostal incision and single large gall bladder stone (6 cm) retrieved, en-block aponeurotic muscle skin and fistula orifice excision done, subtotal cholecystectomy done as CBD, colon, duodenum were plastered together, and further dissection could not possible.

Drain kept in hepatic fossa. During surgery common bile duct, colon, jejunum, stomach inspected carefully for any communication. In post-operative period patient was shifted to intensive care unit. On post-operative day 2 (POD2), the patient had delirium, but serum electrolytes were normal.

So, in view of hepatic encephalopathy serum ammonia levels were done, that were raised (65 µmol/L). Syrup lactulose was given. On POD 4, the patient become stable and shifted to ward and started orally. Drain removed on POD 6. and patient discharged on POD 8. Histopathology reports confirmed the acute on chronic cholecystitis, without any evidence of malignancy.

DISCUSSION

The pathophysiology of the condition involves increased pressure in the gallbladder, secondary to cystic duct obstruction, either caused by a calculus or neoplasia.[2] Predisposing factors are cholelithiasis, infections, malignancy, diabetes, atherosclerosis, prolonged high dose steroids associated disease like polyarteritis nodusa.[3,4] Fundus most distant from cystic artery and physiologically least vascularised therefore most susceptible to ischemia hence is the most common site of perforation.[5,6]

The increase in intraluminal pressure compromises the venous and lymphatic drainage of gallbladder resulting in necrosis and finally gallbladder perforation.[7] Spontaneous biliary fistula can be either internal or external with the majority internal.[8] Internal fistula connections occur at the duodenum 77%, colon 15%, stomach, jejunum, Common bile duct.[9] Generally, fistula or abscess present in right upper quadrant, although other locations such as epigastrium, umbilical, right groin, even the gluteal region, right breast have also been described.[3,10,11] Rare cases of biliary fistulas joined to regions such as the renal pelvis, uterus, vagina, ovary, urinary bladder, hepatic artery, portal vein, pericardium, and bronchial tree have also been reported.[12-14] Perforation of the gallbladder with cholecystohepatic communication is a rare cause of liver abscess.[15]

Niemeier in the 1930s, proposed a classification of gallbladder perforation: Type I - acute free perforation into peritoneal cavity, without protective adhesions; Type II - subacute perforation with pericholecystic abscess; and Type III - chronic perforation with cholecystoenteric fistula.[3,16] A fourth type is suggested by Andersen et al Type IV: cholecystobiliary fistula formation.[17]

Roslyn et al reported in their study that type I and type II gallbladder perforations are mostly seen in young patients (< 50 years), and type 3 is seen in elderly with long history of stone disease.[18] Prior to a spontaneous rupture is a state referred to by Nayman as "empyema necessitatis". The state refers to the abdominal wall featuring a "burrowing abscess" as a consequence of inflammation of the gallbladder.[19]

Gallbladder perforation on CECT will show interruption of the gallbladder wall or a focal defect, gallstone may be noted outside gallbladder and pericholecystic abscess formation may be confined to gall bladder or rarely extend into the liver.[3]

A recommended approach for surgical management of SCF: Two stage surgical treatment for septic patient with poor clinical outcome. External drainage of the abscess and antibiotics were used for enabling biliary drainage and sepsis control then cholecystectomy and fistula excision. Single stage procedure like the one we performed is considered to be the treatment of choice. Single stage laparoscopic recently performed by Kumar.[1,20] Malik et al describe an approach that involves the laparoscopic removal of the gallbladder and dissection but not excision of the fistula from the abdominal wall.[21]

A 40% mortality rate was associated in one study with type 1 free perforation and a lower rate of 4% mortality rate with Type 2.[22] Due to the non-specific clinical signs, medical imaging, particularly CECT, is crucial in distinguishing SCF. Magnetic resonance imaging cholangiopancreatography (MRCP) is also used when CT is inconclusive.[15]

CONCLUSION

Prompt surgical intervention is warranted to decrease morbidity and mortality in patients with gallbladder perforation. Intraoperative inspection of CBD and other viscera and meticulous post-operative monitoring in elderly patient with significant co-morbidity. While SCF is a rare but possible condition, there are a significant number of cases of acute cholecystitis which warrant further practitioner awareness of diagnosis and treatment options.

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