Spontaneous Cholecysto-Cutaneous Fistula Complicating Carcinoma of the Gall Bladder: A Case Report

Kuljinder Sodhi · Mohd Athar · Vijay Kumar ·
Inder D. Sharma · Nuzhat Husain

Received: 31 March 2009 / Accepted: 30 July 2009 / Published online: 16 August 2011
© Association of Surgeons of India 2011

Abstract Most cholecystocutaneous fistulas are postoperative complications of liver and biliary tract surgery or trauma. External biliary fistulas rarely occur spontaneously as a result of intrahepatic abscess (pyogenic or parasitic), necrosis or perforation of the gallbladder, or other inflammatory process involving the biliary tree. A cholecystocutaneous fistula as a presentation of an underlying cancer arising from the gall bladder is an extremely uncommon finding. Over the past 50 years fewer than 20 cases of spontaneous cholecystocutaneous fistulas have been described in the medical literature but so far there has been no published report of a cholecystocutaneous fistula arising from adenocarcinoma of gall bladder. We here report a case of a patient presenting with spontaneous cholecystocutaneous fistula from cancer of gall bladder.

Keywords Cholecystocutaneous fistula · Gall bladder adenocarcinoma · Chemotherapy

Introduction

Gall bladder cancer is an aggressive malignancy with a dismal outcome. Over about three quarters of patients present with cancer beyond the scope of resection secondary to metastatic or locally advanced disease. [1] A cholecystocutaneous fistula as a presentation of an underlying cancer arising from the gall bladder is an extremely uncommon finding. Over the past fifty years fewer than twenty cases of spontaneous cholecystocutaneous fistulas

have been described in the medical literature but so far there has been no published report of a cholecystocutaneous fistula arising from adenocarcinoma of gall bladder. Spontaneous external biliary fistula was first described as early as in 1670.

Most cholecystocutaneous abscesses or fistulas are postoperative complications of liver and biliary tract surgery or trauma. External biliary fistulas rarely occur spontaneously as a result of intrahepatic abscess (pyogenic or parasitic), necrosis or perforation of the gallbladder, or other inflammatory process involving the biliary tree. [2, 3] We here report a case of spontaneous cholecystocutaneous fistula as a presenting feature of a cancer gall bladder

Case History

A 66 year old female presented with the complaint of pain in the right hypochondrium for the last seven months. On clinical examination a large tender abdominal lump with induration on overlying skin was felt in the right upper abdomen. Patient was advised for admission, but she declined. She presented after one week with the complaint of stones coming out of anterior abdominal wall. There was no history of trauma, needle injury or surgical procedure carried out. On examination a small fistulous opening overlying the right upper abdomen with surrounding abdominal wall induration was seen. Small stones were seen coming out from the opening (Fig. 1). Routine blood investigations were under normal limits. Liver function test revealed raised total bilirubin of 3.6 mg/dl, direct bilirubin of 2.0 mg/dl and alkaline phosphatase of 602 IU/L. AST and ALT were 401 IU/L and 560 IU/L respectively. Ultrasonography of abdomen revealed a mass in the gall bladder with multiple gall stones with mild intra-hepatic biliary radical

K. Sodhi (✉) · M. Athar · V. Kumar · I. D. Sharma · N. Husain
King George Medical College,
Lucknow, Uttar Pradesh, India
e-mail: doconco08@yahoo.com

Fig. 1 Fistulous opening in the right upper abdomen showing stone coming out

dilatation with evidence of infiltration of hepatic bed and overlying abdominal wall. An abdominal computed tomography scan showed a mass in the gall bladder with involvement of hepatic bed and infiltration into the overlying abdominal wall along with multiple periportal, peripancreatic and aortocaval nodes (Fig. 2). A fistulogram was carried out using gastrograffin dye injected through the small opening in the anterior abdominal wall; it demonstrated a patent cholecystocutaneous fistula with multiple stones in the gallbladder and the dye reaching the duodenum (Fig. 3). Fine needle aspiration cytology (FNAC) from the gall bladder lump and the infiltrated abdominal wall was done which confirmed the diagnosis of adenocarcinoma gall bladder from both the sites (Fig. 4). She was started on

Fig. 2 CECT upper abdomen showing a gall bladder mass infiltrating anterior abdominal wall

Fig. 3 Fistulogram demonstrating cholecystocutaneous fistula

Gemcitabine (1000 mg/m^2) and cisplatin (70 mg/m^2) based chemotherapy. She had a partial response to the treatment but after 3 cycles she succumbed to the disease within four months of diagnosis

Discussion

Spontaneous external biliary fistula was a known complication of biliary tract disease in the past. This entity was first described as early as in 1670 by Thilesus. However since then less than 20 cases of spontaneous cholecystocutaneous fistula have been reported in last 50 years. [2, 4, 5] So far no case report of a spontaneous malignant cholecystocutaneous fistula has been published. Gall

Fig. 4 FNAC showing adenocarcinoma cells

bladder cancer is an aggressive disease with a dismal prognosis. [1, 6] It has a remarkable propensity to seed and grow in the peritoneal cavity, as well as along the needle biopsy sites and in the laparoscopic port sites. It can also spread early by direct extension into the liver and other adjacent organs.

Spontaneous biliary fistulae are usually a complication of acute suppurative cholecystitis associated with cholelithiasis.[3] The external opening of a cholecystocutaneous fistula is generally in the right hypochondrium. However, other sites can be involved such as the left hypochondrium (45%), the umbilicus (27%), the right lumbar region, the gluteal region [2] and the right iliac fossa.[7] The presence of right upper quadrant mass in association with gall bladder cancer reflects unresectability in more than 90% of the patients. Chemotherapy has been used for palliation of unresectable disease with palliation of symptoms. Chemotherapy seems to have some survival benefit in stage IV disease. [8] The overall outcome of this disease is dismal with the five year survival rate being less than 5% with a median survival of 5 to 8 months. [9]

Spontaneous cholecystocutaneous fistula arising in a case of advanced cancer of gall bladder is an extremely uncommon presentation of this disease. As no such case has been published in the literature, it may be inferred from this case that the disease is in a fairly advanced stage and

beyond surgical salvage at the time when it manifests as a fistula. The intent of treatment is generally palliative and the overall outcome is dismal.

References

1. Bartlett D, Fong Y, Fortner JG, Brennan MF, Blumgart LH (1996) Long-term results after resection for gallbladder cancer. *Ann Surg* 224(5):639–646
2. Nicholson T, Born MW, Garber E (1999) Spontaneous cholecystocutaneous fistula presenting in the gluteal region. *J Clin Gastroenterol* 28(3):276–277
3. Nayman J (1963) Empyema necessitatis of the gall-bladder. *Med J Aust* 1:429–430
4. Vasanth A, Siddiqui A, O'Donnell K (2004) Spontaneous cholecystocutaneous fistula. *South Med J* 97(2):183–185
5. Cubertaond P, Gainant A, Cucchiaro G (1994) Surgical treatment of 724 carcinomas of the gall bladder. Results of the French Surgical Association Survey. *Ann Surg* 219:275–280
6. Gibson TC, Howat JMT (1987) Cholecystocutaneous fistula. *Br J Clin Pract* 41:980–982
7. Flora HS, Bhattacharya S (2001) Spontaneous cholecystocutaneous fistula. *HPB(Oxford)* 3(4):279–280
8. Memon MA, Anwar S, Shiwani MH, Memon B (2005) Gall bladder carcinoma: a retrospective analysis of twenty two years experience of a single teaching hospital. *Int Semin Surg Oncol* 2 (1):6
9. Piehler JM, Crichlow RW (1978) Primary carcinoma of the gall bladder. *Surg Gynecol Obstet* 147(6):929–942