Spontaneous Cholecystocutaneous Fistula: A Case Report

Sibakoti Y.C.*, Basnet RB**, Poudel R***

*Associate Professor & Senior Consultant Surgeon, ** Assistant Professor, *** MS Resident, Department of Surgery, Bir Hospital, NAMS.

ABSTRACT

INTRODUCTION: An external biliary fistula is a rare complication of gallstone disease. We present a case of spontaneous cholecystocutaneous fistula successfully treated with excision, debridement and cholecystectomy in Bir Hospital on July 2012.

CASE OUTLINE: A 60-year-old female presented with a necrotizing fasciitis of anterior abdomen wall, what was thought to be an abscess in the right hypochondrium and was planned for incision, drainage and debridement.

RESULTS: During incision and drainage gall stone was retrieved from the abscess cavity with bile mixed pus which confirmed spontaneous cholecystocutaneous fistula instantly and converted to cholecystectomy under general anesthesia. The fistulous track was excised together with the gallbladder.

DISCUSSION: This condition is rarely seen nowadays due to the greater availability of antibiotic therapy and advanced biliary surgery. Cholecystectomy is the preferred treatment, although in a few patients, the fistula may close spontaneously.

KEY WORDS: external biliary fistula, spontaneous, cholecystocutaneous fistula, gallstone, gall bladder carcinoma.

INTRODUCTION

Biliary fistulae is usually divided into internal and external biliary fistulae. External biliary fistulae can be further subdivided based on etiology into spontaneous, therapeutic, traumatic, and iatrogenic fistulae. Spontaneous external cholecystocutaneous fistula is a very rare surgical complication of neglected gallstone disease. This complication has become even increasingly rarer because of early diagnosis and efficient surgical intervention for gallstone diseases.

Thilesus first described spontaneous cholecystocutaneous fistula in 1670 A.D. Before 1900 A.D, Courvoisier in 1890, Naunyn in 1896 and Bonnet in 1897 published large series of spontaneous cholecystocutaneous fistula in quick succession. Courvoisier reported 169 cases of spontaneous cholecystocutaneous fistula among 499 cases of gallbladder perforation. Naunyn published about 184 cases and Bonnet 122 cases of spontaneous cholecystocutaneous fistula. Since the advent of cholecystectomy for the treatment of gallstone disease, the incidence of spontaneous cholecystocutaneous fistula has reduced dramatically; from 1890-1949, only 37 cases were found in the published literature. A literature review of cases published in the last 50 years reveals fewer than 50 cases. The declining incidence is attributed to prompt diagnosis, availability of antibiotics, and early surgical treatment for calculus cholecystitis and empyema. This decreasing incidence is further confirmed by...
the availability of large series published before 20th century, in contrast to more recent literature, which published only individual case reports (see table no.1).

<table>
<thead>
<tr>
<th>Author(s)</th>
<th>Year Published</th>
<th>Number of Cases</th>
<th>Country of Origin</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gordon et al</td>
<td>2011</td>
<td>1</td>
<td>United States of America</td>
</tr>
<tr>
<td>Sayed et al</td>
<td>2010</td>
<td>1</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>Pezzilli et al</td>
<td>2010</td>
<td>1</td>
<td>Italy</td>
</tr>
<tr>
<td>Metsemakers et al</td>
<td>2010</td>
<td>1</td>
<td>Belgium</td>
</tr>
<tr>
<td>Tallon Aquilar et al</td>
<td>2010</td>
<td>1</td>
<td>Spain</td>
</tr>
<tr>
<td>Hawari et al</td>
<td>2010</td>
<td>1</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>Gandhi et al</td>
<td>2009</td>
<td>1</td>
<td>New Zealand</td>
</tr>
<tr>
<td>Murphy et al</td>
<td>2008</td>
<td>1</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>Ijaz et al</td>
<td>2008</td>
<td>1</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>Chatterjee et al</td>
<td>2007</td>
<td>1</td>
<td>India</td>
</tr>
<tr>
<td>Malik et al</td>
<td>2007</td>
<td>1</td>
<td>United Kingdom</td>
</tr>
<tr>
<td>Nagral et al</td>
<td>2007</td>
<td>1</td>
<td>India</td>
</tr>
<tr>
<td>Marwah et al</td>
<td>2007</td>
<td>1</td>
<td>India</td>
</tr>
</tbody>
</table>

In spontaneous cholecystocutaneous fistula, the abscess is walled off by the abdominal wall and progressively penetrates it. The fistula usually occurs via the fundus of the gallbladder, as this is the farthest from the cystic artery and most likely to be affected by inflammation-induced ischemia. The cholecystic abscess may initially cause a tender inflammatory area or abscess in the abdominal wall and spontaneously rupture, forming a fistula with drainage from the skin.

**Etiology**

This complication is invariably due to a neglected gallstone disease, although isolated case reports have described spontaneous cholecystocutaneous fistula due to carcinoma of the gallbladder and acalculous cholecystitis where there is inflammation in a manner similar to that of gallstones.

*Salmonella typhi*, which has a predilection for the gallbladder, can cause chronic cholecystitis and may predispose the patient to spontaneous cholecystocutaneous fistula. Polyarteritis nodosa with gallbladder vasculitis and steroid use causing immunosuppression also may be associated with the condition.

**Pathophysiology**

The cystic duct or gallbladder is almost always obstructed in patients with spontaneous cholecystocutaneous fistula. In the presence of obstruction, the gallbladder distends and the pressure within rises, impairing the vascular supply. The obstruction and impaired blood supply result in inflammation and may cause focal areas of necrosis. This inflammatory process is typically insidious and recurrent. Surrounding structures wall off the focal area of necrosis. Perforation of the gallbladder may occur, causing a localized cholecystic abscess. In an attempt to discharge this abscess, a fistula may thus form between the gallbladder and stomach, duodenum, colon, or abdominal wall.

In spontaneous cholecystocutaneous fistula, the abscess is walled off by the abdominal wall and progressively penetrates it. The fistula usually occurs via the fundus of the gallbladder, as this is the farthest from the cystic artery and most likely to be affected by inflammation-induced ischemia. The cholecystic abscess may initially cause a tender inflammatory area or abscess in the abdominal wall and spontaneously rupture, forming a fistula with drainage from the skin.

**CASE REPORT**

A 60-year- old -woman presented with painful swelling of the abdominal wall in the right hypochondrium for last 4 days. She also gave history of intermittent pain in right upper quadrant radiating to the tip of shoulder associated with nausea and vomiting since then. She was afebrile and her vital signs were all normal. She had no history of diabetes, hypertension or any other significant illness or surgery in the past. On physical examination the swelling was red in color about 15cm by 10 cm in size extending from right hypochondrium to epigastic and umbilical region and pus discharging from the wound. It was soft, fluctuating and tender on touch with some crepitations. On investigation, CBC, LFT, Renal panel, PT/INR and other investigations were normal except low hemoglobin (8.1 gm%). The presumed diagnosis was necrotizing fasciitis and/ or abdomen wall abscess. She was admitted and treated with intravenous antibiotics covering both aerobic and anaerobic bacteria and was planned for wound debridement and drainage of abscess under intravenous anesthesia (IVA).

**RESULTS**

During incision and drainage thick foul smelling bile stained pus (200ml) was obtained, necrotizing fasciitis of subcutaneous, rectus abdominis muscle was also seen, a gallstone and a fistulae track was found. Under general anesthesia the track was excised and cholecystectomy was performed; the gallbladder was necrosed and perforated at the fundus. Thus the final diagnosis of spontaneous cholecystocutaneous fistula was established. The wound was closed primarily by applying tension sutures and a subhepatic drain was left in situ. There was bile colored discharge from the wound site for a week which stopped spontaneously. The patient made a slow but uncomplicated recovery and was discharged on tenth postoperative day.
Histological examination of the gallbladder showed adenocarcinoma of gallbladder (T₂).

Figure 1. Showing gallstone in the abscess cavity

Figure 2. Showing fistula tract

Figure 3. Tension suture at the end of operation

DISCUSSION

Spontaneous biliary fistula can be either internal or external. Internal fistula are very much commoner, 75% of them connecting to the duodenum and 15% to the colon.¹,²,³ The remaining 10% of internal fistulas connect with the stomach or jejunum, or have multiple communications such as cholecystoduodenocolic fistula.⁴ Spontaneous external biliary fistulas are rare. They are usually due to complication of gallstone disease, but can occur secondary to biliary injury during a surgical procedure, carcinoma gallbladder, cholangiocarcinoma and other traumatic causes.⁵ 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 right iliac fossa and the gluteal region.⁶,⁷

A spontaneous fistula such as this one could be an end result of perforation of the gallbladder secondary to acute or chronic calculous cholecystitis or carcinoma of gallbladder. Perforation of the gallbladder can also occur, rarely, in the absence of gallstones.²

In our case presented here, the likely pathological process was recurrent gallbladder inflammation secondary to gallbladder calculi and carcinoma of gallbladder, causing adherence to the abdominal wall with eventual fistulation, but what is surprising in this case is the lack of severe symptoms experienced by the patient prior to abscess formation. Our patient didn’t know that she has gallstone and/or cholecystitis or cancer gallbladder as she was apparently well until four days before the appearance of painful swelling in right upper abdomen.

In the literature, two cases of combined internal as well as external fistulas have been described, communicating in each case with the duodenum.⁹,¹⁰

The typical presentation of a persistent discharging sinus should suggest the diagnosis, particularly in an elderly patient with a previous history of gallstones or jaundice.¹¹,¹² The management of an external biliary fistula clearly depends on the underlying aetiology.¹³,¹⁴,¹⁵ The acute phase requires treatment with adequate antibiotics, analgesia and resuscitation. In a few proportion of patients the external biliary fistula will heal spontaneously, and therefore operation may be avoided if the patient is elderly or very debilitated.¹⁶ Possible surgical options include cholecystostomy with removal of the gallstones or cholecystectomy. As cholecystostomy carries the possibility of further
Spontaneous cholecystocutaneous fistula is a rare entity in present time where prompt diagnosis and treatment of gallstone disease is very advanced nevertheless fistulae may be at times due to cancer gallbladder.

REFERENCES


