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April 14th, 2011

Dear Sir,

We are submitting a manuscript entitled “**Spontaneous Cholecystocutaneous Fistula Managed with Percutaneous Transhepatic Gallbladder Drainage**” as a Brief Report for consideration for publication in your *Journal*. We present a 21-year-old young lady who developed spontaneous cholecystocutaneous fistula that opened externally at the site of a healed surgical scar. The fistula healed under secondary intension after percutaneous transhepatic gallbladder drainage with uneventful recovery. The manuscript and figures have not been previously published. The manuscript is not under consideration elsewhere. We hope that the manuscript is suitable for publication in your *Journal*.

Sincerely yours,

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Spontaneous Cholecystocutaneous Fistula Managed with Percutaneous Transhepatic Gallbladder Drainage

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Spontaneous cholecystocutaneous fistula is an uncommon complication of gallbladder disease. We herein report a case of spontaneous cholecystocutaneous fistula that opened externally at the site of a healed surgical scar in the mid upper quadrant. The fistula healed under secondary intension after percutaneous transhepatic gallbladder drainage (PTGBD) with uneventful recovery.

A 21-year-old lady was transferred to the emergency department (ED) of this hospital because of a large soft tissue defect at posterior trunk and sacral area after a major trauma. She had been well until 3 weeks earlier when she sustained a motor-vehicle accident which resulted in pelvic fracture with perinorectal laceration; burst fracture at third lumbar spine; 4th and 5th lumbar spine instability and spinal cord compression; sacral fracture and right bimalleolar open fracture. She received emergent surgical intervention, including open reduction and internal fixation for her spinal and lower limb fractures and external fixation for her pelvic fractures, and repair of perinorectal laceration.

Ten days after the trauma, she received elective colostomy because of the poor healing of the rectal wound. However, her postoperative course was complicated with methicillin-resistant *Staphylococcus aureus* (MRSA) wound infection over her back with sepsis and respiratory failure. After removing the implants over her spine and external fixation on her pelvis, there was a huge soft tissue defect over her posterior trunk and sacral area. She then developed ileus and then received total parental nutrition (TPN). Three weeks after the trauma, she was transferred to the ED of this hospital.

Upon transferring to our hospital, the patient appeared critically ill and was under mechanical ventilator support. On examination, she had a distended abdomen and large soft tissue defect over her lower back and buttock. The blood pressure was 134/68 mmHg, the temperature was 38.6°C, and the pulse was 124 beats per minute. The white-cell count was 18,720/ μ l (normal range, 3,990 to

10,390), with 81.5% neutrophils (normal range, 40 to 74%) and the C-reactive protein level was 12.89 mg/dl (normal range, < 0.8). She had a total bilirubin level of 1.81 mg/dl (normal range, 0.2 to 1.3), an alkaline phosphatase level of 209 IU/L (normal range, 38 to 126), and a gamma-glytamyl transpeptidase level of 107 IU/L (normal range, 8 to 50). Results were normal for the tests of liver function. The abdominal computer tomography revealed a distended gallbladder without the presence of stone (Figure 1a).

Two days later, a ruptured lesion at the site of the healed surgical scar with an internal continuation was noted (Figure 1b). The diagnosis of a cholecystocutaneous fistula was confirmed with a fistulogram (Figure 1c). After the use of PTGBD, her ileus improved gradually. Culture from the drained bile grew *Bacteroides fragilis*. Three weeks later the drainage tube was removed and the fistula healed under secondary intention (Figure 1d). The patient had an unremarkable recovery after series of reconstructive surgeries for the wounds over her lower back and buttock and was discharged home after 66 days of admission. The follow-up in the surgery clinic for two years was uneventful.

Description of spontaneous cholecystocutaneous fistula dates back to Thilesus in 1670. It was common in the last century, but has become rare since the advance of diagnostic instruments and improvement of surgical techniques in gallbladder diseases. After the 205-case series reported by Henry and Orr in 1949, spontaneous cholecystocutaneous fistula has appeared only in individual case reports in the literature¹.

Spontaneous cholecystocutaneous fistula is almost always a result of neglected biliary tract disease. It usually presents as an enlarging mass before rupture, most commonly in the right upper quadrant of the abdomen. However, perforation of the gallbladder without stone was mentioned in 0.6 - 1% of all cases with acute cholecystitis. The etiology of perforation among these cases is unclear.

Bacteremia, steroids, typhoid and trauma are regarded as possible causes. The net effect of infection and circulatory changes is to provide the necessary conditions for necrosis and perforation of the relatively avascular fundus².

Management of cholecystocutaneous fistula requires prompt diagnosis, administration of broad-spectrum antibiotics, adequate drainage, and general supportive care. The diagnosis of cholecystocutaneous fistula is confirmed with the fistulogram. It allows visualization of the fistula and its origin. Closure of the cholecystocutaneous fistula requires elimination of the underlying pathology. It is reported that after the acute inflammation has subsided, an elective cholecystectomy and removal of the fistula can be performed³. However, up to 20% of all external biliary fistulae heal spontaneously suggested that it can be treated conservatively. Surgical intervention may be deferred depending on clinical symptoms¹. In our patient, the developed cholecystocutaneous fistula may be the result of a neglected acalculous cholecystitis possibly due to her previous MRSA sepsis and TPN use. After the confirmation of the fistula by the fistulogram, the PTGBD not only functioned as a device for decompression, but also successfully preserved the gallbladder of this young lady.

Reference:

1. Henry CI, Orr TG. Spontaneous external biliary fistulae. *Surgery* 1949, 26: 641-646.
2. Birch BR, Cox SJ. Spontaneous external biliary fistula uncomplicated by gallstones. *Postgrad Med J* 1991;67:391-2.
3. Roslyn J, Busuttill RW. Perforation of the gallbladder: a frequently mismanaged condition. *Am J Surg* 1979;137:307-12.

Legends

Fig 1a. Upon transferring to our hospital, the abdominal computer tomography revealed distended gallbladder.

Fig 1b. The ruptured lesion was at the site of the healed surgical scar with a internal continuation.

Fig 1c. The diagnosis of a cholecystocutaneous fistula was confirmed by the fistulogram.

Fig 1d. The cholecystocutaneous fiatula headed under secondary intention.

Figure(s)
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