Case 4584

BILIO-UMBILICAL FISTULA: A RARE CASE OF SPONTANEOUS EXTERNAL BILIARY FISTULA

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Patient: 78 years, female

Clinical History:

Bilio-umbilical fistula is a rare form of spontaneous external biliary fistula. We present a case of spontaneous external biliary fistula with drainage through the umbilical orifice in a patient who had had no recorded previous episode of acute cholecystitis, although she did have a clinical history of biliary pathology.

Imaging Findings:

We present the case of a 78-year-old woman with multiple acute episodes of probably biliary origin over the last 45 years. The patient came to the hospital because of a further episode of pain in the right hypochondrium during the previous three months and suppuration from the umbilicus for two months. Furthermore, the patient described the expulsion of “green stones” from the umbilicus. An abdominal echography was performed in which the gall bladder was not clearly identified; the gallbladder fossa was taken up by an acoustic shadow suggestive of a “stone-filled gallbladder”. From this area and extending to the abdominal wall as far as the umbilicus, there was a fusiform hypo-anechoic image in the centre of which there were hyperechogenic images with a posterior acoustic shadow (Figure 1). With a suspicion of a fistulous bilio-umbilical tract, we carried out a CT scan and CT fistulography. The presence of a thick-walled collection with lithiasis and gas inside was confirmed. It was below the theoretical site of the gallbladder and continued with a linear trajectory that traversed the abdominal wall and terminated in the umbilical orifice (Figures 2A-2B). When iodinated contrast was introduced through the umbilicus, the whole fistulous trajectory and the collection were filled, confirming the initial suspicion of a spontaneous bilio-umbilical fistula (Figures 2C, 2D and 2E). The patient was programmed for a surgical intervention but did not turn up.

Discussion:

Biliary fistulae are classified as external (bilio-cutaneous) or internal (e.g. bilio-biliary, bilio-enteric). The majority of internal fistulae are spontaneous, while the external ones are generally secondary to surgery or percutaneous interventions (1). Spontaneous cholecystocutaneous fistula is a rare complication of biliary disease. It usually occurs after repeated attacks of cholecystitis, which lead to adhesion of the gallbladder to the abdominal wall and rupture. It reflects biliary disease that has not been adequately treated (2, 3, 4, 7, 8, 9, 10). Cholecystocutaneous fistula is more frequent in women and often seen in the elderly, in whom surgery entails more risk and cerebral arteriosclerosis may hide the symptoms (4). If the patients are carefully questioned, they characteristically report a long history of biliary disorder. Nevertheless, it should be emphasized that this complication may not be preceded by a clear episode of cholecystitis that would presumably have made the patient seek prompt medical attention (8). The first description of a case of spontaneous cholecystocutaneous fistula is attributed Thilesus in 1670 (3, 4, 5, 7, 8,9).
Curvôisier in 1890 published 499 cases of perforation of the gallbladder, of which a third had formed cutaneous tracts (2, 3, 4, 7,9). Naunyn in his treatise on cholelithiasis published in 1896 says that the most frequent site for opening of the fistula is the periumbilical region, due to the presence of the "umbilical tract", a canal previously occupied by the umbilical vein (8). This is a result of the complex embryological role of the umbilicus (6). In certain cases the fistula is guided by the falci-form ligament to the umbilical region in the midline (5). In general, there is only one spontaneous bilo-cutaneous fistula and in half the cases it opens into the upper right quadrant of the abdomen, although the cutaneous orifice has been observed in other locations as distant as the umbilicus, the left margin of the ribs, the right groin, the thoracic wall or the back (2, 3, 4,8). Umbilical discharge or suppuration is a symptom that is associated with a wide variety of disorders both of embryonic origin (e.g. incomplete regression of the omphalomesenteric tract), and acquired (e.g. Crohn’s disease) (6). The low mortality and morbidity index associated with early surgical treatment of gallbladder lithiasis has lead to a decrease in the incidence of this rare disorder. Thus, since 1900 there has been only occasional mention of this problem, whilst before that date there were many bibliographic references on cholecystocutaneous fistulae (8,9).

**Differential Diagnosis List:** Spontaneous bilo-umbilical fistula in chronic cholecystitis

**Final Diagnosis:** Spontaneous bilo-umbilical fistula in chronic cholecystitis

**References:**


Description: Gray-scale sonogram demonstrates a hypoechoic subcutaneous tract (black arrowheads) going from the theoretical gallbladder fossa (GB), with lithiasis inside (black arrow). Origin:
Description: A. Tomographic section corresponding to the formation of the gas-filled collection (white arrowhead) Origin:
Description: B. The bilio-umbilical fistula track can be seen in the anterior abdominal wall leading from the above-mentioned collection. (thick white arrows) **Origin:**

Description: C. Fistulography- CT. Filling of the chronic collection after the introduction of contrast through the cutaneous orifice of the umbilicus. (white arrowhead) **Origin:**
Description: D. Fistulography-CT. Retrograde filling of the fistulous tract in the abdominal wall. (black arrows) Origin:

Description: E. Fistulography-CT. Visualisation of the theoretical cystic duct with contrast. (white arrows) Origin: