

# A rare case of Cholecystoduodenal and Cholecystocolonic Fistulae in a Young Girl: A Case Report

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## ABSTRACT

Bilioenteric fistulae are late complications of gall bladder disease. Cholecystoduodenal fistulae are most common, followed by cholecystocolonic fistulae. Cholecystogastric fistulae are the least common of all. The majority of the fistulae result from repeated attacks of acute cholecystitis due to gallstones. In 2% of cases, these fistulae are associated with gall bladder cancer. Co-occurrence of cholecystoduodenal and cholecystocolonic fistula (CCF) is rare. Numerous composite fistulae are even rarer. These conditions have variable clinical presentations. It is hard to diagnose these fistulae preoperatively, despite the latest imaging techniques, and they are found intraoperatively. Hence, this condition is a unique surgical entity, being atypical, rare and complex. We present a rare case of combined cholecystoduodenal and cholecystocolonic fistulae encountered by us in a fourteen-year-old girl, also a known patient of sickle cell anemia. Few such cases have been previously reported.

**KEYWORDS:** Cholecystoduodenal, Cholecystocolonic, Bilioenteric, CCF, BEF, Fistulae

## INTRODUCTION

Bilioenteric fistulae are a rare complication of cholelithiasis<sup>1</sup>. They have a high incidence of morbidity and mortality in older age groups. They may be associated with biliary ileus and may cause about 5% of bowel obstructions<sup>2</sup>. Up to 80% of these cases are cholecystoduodenal<sup>3</sup>. Cholecystocolonic and cholecystogastric fistulae occur in up to 20% of cases<sup>4</sup>. Others of these fistulae are rare. Gallstones lead to repeated attacks of acute cholecystitis and might result in 60% of cases of these fistulae<sup>5</sup>. Calculi cause pressure injury and sloughing of the wall of the biliary tract into any neighboring organ, which in turn occurs due to repeated inflammation of the biliary tract wall<sup>6</sup>. But malignancy, peptic ulcer, trauma, or previous surgery can also cause these fistulae<sup>6</sup>. The co-existence of cholecystoduodenal and cholecystocolonic fistulae is an extremely rare observation.

The symptoms may consist of episodes of abdominal pain, diarrhea, malabsorption and vomiting in cholecystocolonic fistulae<sup>7</sup>. Women are more affected than men, and this is most common between the ages of 60 and 70<sup>8</sup>. The triad of chronic diarrhea, vitamin K malabsorption, and pneumobilia (air in the biliary tract) is pathognomonic for this type of fistula<sup>6</sup>.

Laboratory tests are usually not helpful in the

diagnosis of these fistulae. There might only be an increase in liver cell damage and cholestasis<sup>9</sup>. Only a preoperative diagnosis can help to reduce the high rate of morbidity and mortality related to these fistulae, but it is rarely made. Ultrasound, X-rays, CT and MRI can, however, help make a preoperative diagnosis of these fistulae. Multiple or composite fistulae are rare and have been reported in only a few cases<sup>7</sup>.

## CASE REPORT

A fourteen-year-old girl was brought to the hospital with a history of pain in the upper right part of the abdomen, with jaundice, nausea and vomiting for two days. She had sickle cell disease and had been previously admitted to the hospital for a sickle cell crisis. She also had many blood transfusions for this condition. WBCs ( $17.3 \times 10^3$ ) and total and direct bilirubin (39.74 and 5.13) were significantly elevated. Other blood reports revealed low hemoglobin (9.6 g/dl), but serum sodium, potassium, blood urea nitrogen, creatinine, aspartate transaminase, alanine aminotransferase, alkaline phosphatase, INR, PTT and PT were normal. Ultrasound showed a partially contracted gallbladder with a diffusely thickened wall measuring 7 mm. There was severe peri-cholecystic edema present. There were multiple stones inside the gall bladder. One stone of diameter 13 mm was fixed in the gall bladder neck. There was a gas shadow at the fundus of the gallbladder and the surrounding tissues. The gallbladder was tender. CBD had multiple stones and was dilated. She was admitted as a case of acute cholecystitis with obstructive jaundice. She underwent ERCP. CBD was cleared of stones, and a stent was left in the CBD. She left the hospital with a plan for laparoscopic cholecystectomy later. She presented with obstructive jaundice after four weeks. Both total and direct bilirubin were high, and

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ultrasound showed a dilated CBD with multiple stones. She had to undergo a repeat ERCP. There was a stricture at the Ampulla of Vater. A sphincterotomy was performed, and the CBD was cleared of the stent and stones.

After the insertion of a plastic stent, the patient's symptoms improved, as did the lab results. She was discharged with a plan for elective cholecystectomy. Before surgery, her lab reports were normal. On laparoscopic surgery, the transverse colon and duodenum were found to be strongly stuck to the gall bladder. One fistula was present between the gall bladder fundus and transverse colon, and another one was present between the gall bladder body and duodenum. During blunt dissection, the cholecystoduodenal fistula opened up. An upper midline incision was given for open surgery. Repair of the colon and duodenum was done. Cholecystectomy was also performed. The patient recovered smoothly and was discharged on the fourth postoperative day. Two months later, the CBD stent was extracted during endoscopy. Patient follow-up was performed for 1 year, and no complications were found.

## DISCUSSION

Most of the bilioenteric fistulae result from recurrent inflammation of the gallbladder (cholecystitis)<sup>9</sup>. Cancer, amebic infections, trauma, peptic ulcers, and diverticulitis can also result in these fistulae<sup>10</sup>. In this particular patient, the cause of these fistulae was recurrent attacks of cholecystitis. Mostly these fistulae are a late outcome of gallstone disease. They are usually found in the sixth and seventh decades of life<sup>8</sup>. Our patient was a young girl of fourteen years of age, with both cholecystocolonic and cholecystoduodenal fistulae. No case of a teenager with this cholecystoenteric fistula has been reported in the literature. Most of the bilioenteric fistulae are single. Multiple or complex cholecystoenteric fistula is an uncommon complication of gallstones<sup>11</sup>. In our patient, there were two fistulae: one from the fundus of the gall bladder to the colon, and the second from the body of the gall bladder to the first part of the duodenum.

The typical clinical picture of CCF is a triad of symptoms of diarrhea, right hypochondrium pain, and cholangitis, reflected by jaundice and/or fever<sup>12</sup>. It has a laxative effect on the colon due to bile acids that reach the colon directly, and causes diarrhea<sup>7</sup>; this may also result in a malabsorptive syndrome leading to megaloblastic anaemia, ventricular tachycardia, heart failure, osteomalacia, and fractures of the pelvis<sup>13</sup>. Our patient, although young, fourteen years old, with a weight of only 42 kg, did not give any history of chronic diarrhea, however, there was a history of repeated episodes of acute cholecystitis and sickle cell crisis.

Laboratory tests usually do not help in the diagnosis of these fistulae. They may only show increased liver cell damage and cholestasis; hence, there is a need of further investigation by imaging techniques is

needed<sup>8</sup>. In our patient, liver function tests were normal; however, both direct and indirect bilirubin levels were elevated. This could have been due to her sickle cell anemia and multiple sickling crises.

Even with the latest imaging techniques, preoperative diagnosis remains difficult and is at best between 8-17%<sup>14</sup>. Physicians should consider this diagnosis in patients presenting with recurrent episodes of cholecystitis. Given this clinical picture and ultrasound report, CT/MRCP should be performed before surgery. Most cases are diagnosed intraoperatively, presenting difficulty for the surgeons who have to change the elective cholecystectomy. Pneumobilia on plain radiography, which was stressed for a long time as a gold standard for diagnosis of these fistulae, is usually not present<sup>13</sup>. Ultrasound, considered an initial and one of the most useful and reliable tools in hepatobiliary conditions, rarely provides useful information about these fistulae<sup>12</sup>. Ultrasound is used to make a diagnosis of chronic cholecystitis and is useful for full screening of the abdomen. Barium enema sometimes leads to preoperative diagnoses of cholecystocolonic fistula but has low sensitivity (high false-negative results).<sup>13</sup> Contrast imaging techniques are advocated to detect air in the biliary tract and suggest a fistula, although their significance in detecting gallstones and ruling out an associated malignancy is greater. ERCP, colonoscopy and endoscopic ultrasound detect cholecystocolonic fistulae (CCF) in occasional cases, but their results are very inconsistent<sup>1</sup>. Because they are invasive, these investigations are usually not performed in patients with nonspecific symptoms.

In the very few cases of preoperative diagnosis of this fistula, several tests have been performed. The initial ultrasound, performed before ERCP, and the follow-up ultrasound showed gas in the gallbladder fundus, which should have raised suspicion of a bilioenteric fistula. If this sign had been recognised and followed up with MRCP or CT, the diagnosis of the fistula could have been made preoperatively. Hence, given this clinical picture and ultrasound report, CT/MRCP should be performed before surgery.

The approved treatment of bilioenteric fistulae is cholecystectomy with repair of the fistula<sup>7</sup>. The failure to diagnose it preoperatively results in a more complex procedure due to the unexpected scenario. The procedure might need breaking of adhesions and resection of bowel. Various approaches have been reported for their treatment, from ERCP and sphincterotomy alone to major gut resection. Surgery has to be tailored according to the patient. Many authors support laparoscopic surgery, whereas many of them report that the laparoscopic approach causes long operating times and leads to conversion eventually<sup>15</sup>.

In our patient, surgery was started as an elective laparoscopic cholecystectomy. While separating adhesions between the gallbladder, colon, and duodenum, the fistulous tracks inadvertently opened.

Hence, the procedure had to be converted to an open one due to a lack of experience with laparoscopy in such cases. An open cholecystectomy was performed, with the excision of the tracks of these fistulae, and repair of the colon and duodenum. Both the small and large bowel were examined for any embedded stones.

### CONCLUSION

Only a high suspicion and repeated examinations disclose a fistula. In this case, despite a tell-tale sign of gas in the gallbladder fundus on ultrasound, this finding was not given importance.

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