CASE REPORT

Gallstone pancreatitis secondary to a pathological cystic duct stump in a setting of chronic pain syndrome: a diagnostic dilemma

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SUMMARY

A 32-year-old woman presented with epigastric pain. She was a patient with chronic pain syndrome and had visited the emergency department several times over the past year. She did not drink alcohol. She had a laparoscopic cholecystectomy for gallstone pancreatitis 4 years ago. Her lipase was 2020 with normal bilirubin. MR cholangiopancreatography revealed a cystic structure resembling a gallbladder in the gallbladder fossa. This was in connection with the biliary system. The structure also contained stones. A review of the histology did confirm the gallbladder had been removed. She proceeded to have a laparoscopic re-cholecystectomy and made an unremarkable recovery.

BACKGROUND

This case presents a rare cause for pancreatitis. It also highlights the potential diagnostic difficulty in patients with chronic pain.

CASE PRESENTATION

A 32-year-old woman of Maori ethnicity was admitted with a 6-day history of epigastric pain radiating to her back. This was associated with nausea and vomiting. She has had several previous episodes over the past year which seemed to be aggravated by fatty foods. She did not drink alcohol and her bowels were normal with no blood. She had been sent home from the emergency department on several occasions in the past 2 years, with the pain being attributed to her chronic pain syndrome. Several investigations had been organised as an outpatient, including an ultrasound, CT abdomen and pelvis, gastroscopy and colonoscopy all of which were normal and performed between 1 and 2 years ago.

She had undergone a laparoscopic cholecystectomy in 2010 for gallstone pancreatitis. According to the operative note, the surgical procedure was uncomplicated involving removal of the thin, walled gallbladder. An intraoperative cholangiogram (IOC) showed normal anatomy. The cystic duct was clipped. The patient had an uncomplicated recovery.

With regard to her background, she was seeing a pain specialist for a 10-year history of chronic abdominal pain although she states that this pain was different. She has a history of endometriosis, obesity, polycystic ovarian syndrome and asthma.

On examination she was noted to be morbidly obese. There were no signs of jaundice. Her

abdomen revealed port site scars consistent with a laparoscopic cholecystectomy. She was tender to palpation in the epigastrium/right upper quadrant but there was no peritonism.

INVESTIGATIONS

Blood revealed a normal full blood count. Her renal function was normal. However, her lipase was 2020 (normal range: 13–60), bilirubin was 6 (normal range: 0–24), γ -glutamyl transpeptidase of 92 (normal range: 0–50), alanine transaminase and aspartate aminotransferase of 57 (normal range: 0–45). Her lipids and calcium were normal.

She proceeded to have an abdominal ultrasound scan which found a 3×2×3 cm cystic structure in the gallbladder fossa. A subsequent MR cholangiopancreatography (MRCP) confirmed a cystic structure resembling a gallbladder in the gallbladder fossa. This connected with the biliary system and also contained stones. The previous clip was close by or part of this complex.

DIFFERENTIAL DIAGNOSIS

Given the markedly elevated lipase and clinical history of upper abdominal pain, the diagnosis of pancreatitis was made. In context of the history, laboratory and imaging findings, this was secondary to gallstones. The Modified Glasgow Score was consistent for mild pancreatitis.

TREATMENT

She underwent laparoscopic re-cholecystectomy during the admission. There were multiple adhesions around the gallbladder fossa. After careful dissection and mobilisation of the cystic structure, it was excised at the gallbladder-cystic duct junction leaving a cuff on the cystic duct which was oversewn laparoscopically. Figure 1 is an intraoperative picture of the opened pathological cystic duct stump (CDS). The mass contained several small stones. Histological features were consistent with chronic cholecystitis of a gallbladder type structure.

OUTCOME AND FOLLOW-UP

The patient recovered well from surgery and was discharged home 2 days postoperatively.

DISCUSSION

Laparoscopic cholecystectomy remains the best practice operation for managing cholelithiasis in the modern world, achieving satisfactory outcomes in the majority of patients.

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Figure 1 Intraoperative view with pathological cystic duct stump opened.

Post-cholecystectomy syndrome (PCS) has a varied reported incidence of 10–50%.^{1–6} The wide variation may be secondary to patients with recurrent extra biliary type symptoms. This leaves a small minority of patients with PCS with a true biliary cause, such as biliary strictures, retained biliary colic, cystic duct stump syndrome/retained gallbladder remnant, and stenosis or dyskinesia of the sphincter of Oddi.^{7–10}

Our case highlights the need to consider the possibility of a pathological cystic duct stump or retained gallbladder remnant when dealing with PCS, especially in the setting of a chronic pain syndrome, as in our patient.

The ultrasound and MRCP findings of a cystic structure in the gallbladder fossa of a patient postcholecystectomy can be secondary to gallbladder duplication, a type II choledochal cyst or a gallbladder remnant/secondary dilation of a CDS.

The nature of the cystic structure in our patient is highly unlikely to be attributed to gallbladder duplication because this patient had previously normal preoperative ultrasound as well as a normal IOC.

The most popular definition of a gallbladder remnant has been attributed to Bodvall *et al*¹¹ in which it is defined as ‘a wider part of the free end of the remnant cystic duct, giving the impression of a diminutive gallbladder’. Interestingly, Bodvall *et al*¹¹ also found that a CDS longer than 1 cm was present in 67% of patients with common bile duct stones and 82% of patients with severe postoperative biliary symptoms.

It can be difficult to differentiate between a gallbladder remnant and a grossly dilated CDS macroscopically and even microscopically,¹² the origins and pathophysiology of both needs to be better understood.

Reviewing the operative note from the initial laparoscopic cholecystectomy, the cystic structure, as seen in our patient, is probably a grossly dilated long CDS which has since become symptomatic. Selvaggi *et al*¹³ hypothesised that a CDS may be grossly dilated secondary to chronic obstruction associated with biliary sludge that collects in a low pressure structure like a cystic duct, which over time may progress to actual calculi. These calculi can also be associated with further inflammation and/or strictures within the cystic duct. The possibility of missing retained cystic duct calculi on the intraoperative cholangiogram during the initial cholecystectomy must also be taken note of.

Keiler *et al*¹⁴ found that 65% of 113 post-cholecystectomy patients who underwent intravenous cholangiography had a >1 cm cystic duct present. The incidence of a long CDS is said to be higher with a laparoscopic procedure compared to open² because the cystic duct is usually clipped closer to the common

bile duct (CBD) in an open procedure. Therefore, some papers^{15 16} have recommended that the cystic duct should be divided closer to the CBD than is routinely performed nowadays, ideally leaving a CDS no longer than 0.5 cm. However, this may potentially lead to more CBD injuries.

Rogy *et al*⁷ looked at the significance of a CDS found in 35 of 322 patients who underwent a second operation of the biliary system. They concluded that having a long CDS (>1.5 cm) is not pathological unless associated with a secondary pathology like retained calculi.

Patients with PCS secondary to a retained gallbladder remnant or a CDS may present at variable intervals. Mergener *et al*¹² described a patient who was diagnosed 25 years after initial cholecystectomy, whereas Walsh *et al*¹⁵ in their series reported a range of 14 months to 20 years to diagnosis from initial cholecystectomy. Unfortunately, our patient had various presentations with abdominal pain and occasionally mildly elevated liver function tests. This was investigated thoroughly, as described above, but tests did not reveal a cause for her symptoms. The complicated nature of her medical history may have contributed to the delay in her diagnosis.

Nevertheless, an ultrasound of the abdomen as well as obtaining a set of routine laboratory workup is the first step in the investigation of PCS. As in the case of our patient, ultrasound examination may not be sensitive and specific for a gallbladder remnant or CDS unless it is of a large size or contains calculi within it.¹⁷ Following that, either a MRCP or endoscopic ultrasound would be warranted if symptoms highly suspicious for PCS persist.^{13 17–19} ERCP can then be performed if therapeutically indicated.

Once the diagnosis of either a gallbladder remnant or pathological CDS is established, a re-cholecystectomy is the definitive treatment modality.^{13 17 19} From our literature search, we could not identify a general consensus on the ideal approach for re-cholecystectomy. There were equal arguments favouring either an open or laparoscopic approach, with the laparoscopic approach favoured in advanced laparoscopic centres. The laparoscopic approach chosen in our patient was difficult but achievable with careful dissection.

Learning points

A pathological cystic duct stump (CDS) can be a diagnostic dilemma especially in the setting of chronic pain. Ultrasound of the abdomen may not be as sensitive or specific for a pathological CDS; therefore, further imaging modality with MR cholangiopancreatography or endoscopic ultrasound may be warranted to prevent a delay in diagnosis. Thereafter, laparoscopic re-cholecystectomy can be a safe and definitive option for management of a pathological CDS.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Lasson A. The postcholecystectomy syndrome: diagnostic and therapeutic strategy. *Scand J gastroenterol* 1987;22:897–902.
- 2 Lum YW, House MG, Hayanga AJ, *et al*. Post cholecystectomy syndrome in the laparoscopic era. *J Laparoendosc Adv Surg Tech A* 2006;16:482–5.

- 3 Tsoraides SS, Cha AI, Crawford D. Postcholecystectomy biliary symptoms. *J Surg Educ* 2007;64:228–33.
- 4 Venu RP, Greenen JE. Postcholecystectomy syndrome. In: Yamada T, ed. *Textbook of gastroenterology*. 2nd edn. Philadelphia: Lippincott, 1995:2265–77.
- 5 Christiansen J, Schmidt A. The Postcholecystectomy syndrome. *Acta Chir Scand* 1971;137:789–93.
- 6 Ekdahl PH. On late distress following biliary tract operations. *Acta Chir Scand* 1953;106:339–50.
- 7 Rogy MA, Fugger R, Schulz F. Reoperation after cholecystectomy. The role of the cystic duct stump. *HPB Surg* 1991;4:129–34; discussion 134–5.
- 8 Bar-Meir S, Halpern Z, Bardan E, et al. Frequency of papillary dysfunction among cholecystectomized patients. *Hepatology* 1984;4:328–30.
- 9 Stefanini P, Carboni M, De Barnardinis G. Factors influencing the long term results of cholecystectomy. *Surg Gynaecol Obstet* 1974;139:734–8.
- 10 Glenn F, Cameron JL. Complications following operations upon the biliary tract and their management. In: Hardy JD, ed. *Complications in surgery and their management*. Philadelphia: WB Saunders, 1981:512–18.
- 11 Bodvall B, Overgaard B. Cystic duct remnant after cholecystectomy: incidence studied by by cholegraphy in 500 cases, and significance in 103 reoperations. *Ann Surg* 1966;163:382–90.
- 12 Mergener K, Clavien PA, Branch MS, et al. A stone in a grossly dilated cystic duct stump: a rare cause of postcholecystectomy pain. *Am J Gastroenterol* 1999;94:229–31.
- 13 Selvaggi F, Di Bartolomeo N, De Iuliis I, et al. Laparoscopic treated so-called reformed gallbladder in patient with postcholecystectomy chronic pain. *G Chir* 2011;32:335–7.
- 14 Keiler A, Pernegger C, Hornof R, et al. The cystic duct stump after laparoscopic cholecystectomy. *Wien Klin Wochenschr* 1992;104:356–9.
- 15 Walsh RM, Ponsky JL, Dumot J. Retained gallbladder/cystic duct remnant calculi as a cause of postcholecystectomy pain. *Surg Endosc* 2002;16:981–4.
- 16 Hinkel CL, Miller GA. Correlation of symptoms, age, sex, and habitus with cholecystographic findings in 1000 consecutive examinations. *Gastroenterology* 1957;32:807–15.
- 17 Hussain M, Nagral S. Biliary pancreatitis secondary to stones from a gallbladder remnant. *Trop Gastroenterol* 2010;31:230–3.
- 18 Topiazan M, Salem RR, Robert ME. Painful cystic duct remnant diagnosed by endoscopic ultrasound. *Am J Gastroenterol* 2005;100:491–5.
- 19 Pernice LM, Andreoli F. Laparoscopic treatment of stone recurrence in a gallbladder remnant: report of an additional case and literature review. *J Gastrointest Surg* 2009;13:2084–91.

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