CASE REPORT

Concurrent cholecystoduodenal fistula and primary aortoenteric fistula

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Abstract

Bilioenteric fistulae are a rare complication and can pose a diagnostic challenge owing to non-specific symptomology. When occurring with an aortoenteric fistula, it represents a rare and potentially life-threatening disease state. We present the case of a 77-year-old gentleman initially treated as presumed ascending cholangitis. This was complicated by upper gastrointestinal bleeding secondary to an aortoenteric fistula and cholecystoduodenal fistula.

INTRODUCTION

A cholecystoduodenal fistula (CDF) is the most common type of bilioenteric fistula, a pathological communication between the biliary system and the gastrointestinal tract. A CDF is specifically a connection between the gallbladder and duodenum, caused by gallstones, surgery, duodenal ulcers, malignant tumours and inflammatory bowel disease [1–3]. The underlying pathophysiology differs between aetiologies. Most commonly, it is caused by chronic inflammation of the gall bladder leading to inflammatory adhesions to the adjacent duodenum, and subsequently eroded through by gallstone [4]. Owing to varied symptomology, the diagnosis of CDF can be challenging.

The incidence of bilioenteric fistulas reported in the literature is 0.9% in the general population and 3.2% in patients diagnosed with biliary disease [5]. The most common symptoms are epigastric pain and sepsis [6, 7]. Patients with bilioenteric fistulae can also present with either obstructive or non-obstructive symptoms. Non-obstructive symptoms include recurrent cholangitis and malabsorption, whilst obstructive symptoms include gallstone ileus, hematemesis or melena [8].

Another rare diagnosis is primary aortoenteric fistulas, which present with massive gastrointestinal bleeding. These occur in the absence of previous endovascular surgery and arise when inflammation of the aortic wall erodes into adjacent bowel. The most commonly cited aetiology is mycotic aortic aneurysms, as a result of bacterial seeding [9, 10].

CASE REPORT

A 77-year-old male was admitted to hospital for anorexia and deranged liver enzymes. He had no significant past medical history, no significant risk factors for liver disease and does not take regular medications. He denied abdominal pain and altered bowel habits. On presentation, he was afebrile, hemodynamically stable and had an unremarkable abdominal examination. However, his blood results showed cholestatic liver enzymes, with an elevated alkaline phosphatase (ALP) of 145 unit/L (30–110), gamma-glutamyl transferase (GGT) of 107 units/L (<55) and total bilirubin of 48 μmol/L (<20). He also had raised a white blood cell count and neutrophil count of 23.7 × 10⁹/L.
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After fourteen inpatient days for monitoring and discharge planning, he became febrile and had worsening ALP and GGT, at 849 and 624 unit/L, respectively. His blood cultures showed Escherichia Coli and Klebsiella pneumoniae bacteraemia. An initial diagnostic computed tomography (CT) scan of the abdomen revealed several hypodense focal liver lesions, multiple thrombosed portal vein branches, pneumobilia in the liver’s left lobe (Fig. 1) and an incidental finding of a 5.8 cm infra-renal abdominal aortic aneurysm (AAA).

He was commenced on Piperacillin–Tazobactam 4.5 g for sepsis from presumed ascending cholangitis. Rivaroxaban 15 mg twice daily was also started for new diagnosis of portal vein thrombosis. The patient initially responded well to medical therapy. However, two weeks later he had a large volume hematemesis and became clinically unstable. A repeat CT abdomen revealed interval increase in the size of the infra-renal AAA to 8 cm (Fig. 2), raising the possibility of impending rupture and aortoenteric fistula.

Rivaroxaban was ceased. An urgent gastroscopy revealed large blood clots in the gastric fundus and body, with a pulsatile lesion at the second part of the duodenum (D2), which was suspicious for an aortoenteric fistula (Fig. 3).

The patient underwent an emergency endovascular aneurysm repair to repair the aortoenteric fistula and admitted to the intensive care unit (ICU) postoperatively.

One-week post-surgery, the patient complained of acute right lower limb pain and swelling. A Doppler ultrasound and CT abdomen angiogram revealed an extensive deep vein thrombosis in the right leg extending to the distal inferior vena cava.

Senior staff deliberated the risks of anticoagulation and ultimately decided to commence an intravenous heparin infusion. Unfortunately, he experienced a second episode of hematemesis five days after despite remaining within therapeutic levels.

A third gastroscopy revealed a bleeding duodenal fistula at the D2/3 junction communicating with the gallbladder, confirming a diagnosis of CDF (Fig. 4). A second laparotomy identified the fistula between the gallbladder to the lateral wall of the duodenum (D2), several stones and sludge in a contracted gallbladder and 6 mm stone in the ampulla. The fistula and the adjacent duodenal wall were repaired, and a cholecystectomy performed.

In the next 24 hours, he had further hematemesis, following which a third laparotomy was performed. Intraoperative findings revealed a deep ulcer in D3. This ulcer formed another aortoenteric fistula with D3, with the aneurysm sac on this occasion bleeding into the duodenum from the iliolumbar arteries and inferior mesenteric artery. The aneurysm sac was opened, and the iliolumbar arteries and inferior mesenteric artery were closed from inside the sac.

The patient survived the surgeries, was admitted into ICU and later discharged from hospital with outpatient follow-up from subspecialty teams. Unfortunately, he was re-admitted 3 weeks later and passed away from an unrelated event.
DISCUSSION
This case highlights the difficulty of diagnosing a biloenteric fistula owing to the non-specific presentation, which could be explained by co-existing pathologies. Compounding this, our patient presented with symptoms found in both obstructive and non-obstructive patterns of presentation with CDF. Given the low incidence rates and faced with another rare diagnosis in the aortoenteric fistula, a biloenteric fistula was not considered in the first instance.

The assumed series of events was that the initial sepsis from E. Coli and K. pneumoniae noted on blood cultures are causes of cholangitis. The severity of the sepsis resulted in the complication of vascular thrombosis. A pre-existing AAA became mycotic and subsequently eroded into the duodenal wall within the D2 and D3 regions, leading to gastrointestinal bleeding concurrent with anticoagulation. The additional findings of a CDF were also notable, a rare complication of cholangitis and chronic gallstones. In retrospect, in the absence of previous biliary instrumentation, persisting pneumobilia on repeat imaging should have raised suspicion for CDF in this patient. Other reasons for pneumobilia include an incompetent sphincter of Oddi or infections of the biliary tract or liver. Endoscopic retrograde cholangiopancreatography may have been useful to define the anatomy and even detect enteric fistulous connections. In addition, there is a potential therapeutic role through stent placement and nasobiliary drainage [8, 11].

This case effectively demonstrates how the combination of non-specific symptomology and multiple rare co-pathologies can lead to delayed diagnosis and management. It emphasizes the importance of considering rare complications of common pathologies, particularly if the clinical course does not progress as anticipated.

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CONFLICT OF INTEREST STATEMENT
None declared.

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ETHICAL APPROVAL AND CONSENT TO PARTICIPATE
This case report did not require review by the relevant Ethics Committees.

CONSENT FOR PUBLICATION
The patient is deceased so obtaining consent was not possible. All patient identifiers and unnecessary information have been removed to protect patient confidentiality.

GUARANTOR
Mohammadali Zad.

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