Bouweret’s syndrome with cholecysto-colic fistula

To the Editor: A 69-year-old man presented to an emergency department with upper abdominal pain, nausea and recurrent non-bilious vomiting of 2 weeks’ duration. The pain was mainly in the epigastric and peri-umbilical regions, and was non-radiating and colicky. There was no history of haematemesis. He also complained of anorexia and weight loss (10 kg). His bowel habits had not changed, and there had been no change in the colour of the stool or urine. Past medical history was unremarkable apart from controlled hypertension and osteo-arthritis.

Physical examination revealed tachycardia, a normal blood pressure and signs of dehydration. There was no evidence of jaundice. The upper abdomen was tender, but there were no palpable masses. Blood tests showed hypokalaemia (2.9 mmol/l), hypochloraemia (87 mmol/l), raised C-reactive protein (28 mg/l), raised gamma-glutamyl transpeptidase (92 U/l) and raised urea (11.4 mmol/l). Results of the remainder of the blood tests were unremarkable. An abdominal radiograph suggested air in the biliary tree. A computed tomography (CT) scan supported the latter finding and confirmed the presence of a large calculus in the proximal duodenum, raising the possibility of a biliary fistula.

Upper gastro-intestinal endoscopy revealed retained fluid and food in the stomach, and a large calculus was clearly visible inside the pylorus, which prevented passage of the scope to the duodenum. During endoscopy a soft guidewire (Jag wire) and balloon were used to obtain access to the duodenum under X-ray screening. Injected contrast filled the duodenum and colon, confirming the presence of another fistula between the gallbladder and the colon. Our attempts to dislodge and remove the calculus endoscopically were futile because it was severely impacted, so laparotomy was carried out.

The colon, which overlaid the area, was separated from the gallbladder revealing a defect in the colon. A large 7×5.5 cm calculus was extracted from the duodenum through a large oblique duodenotomy. Another large 3.3×2.8 cm calculus was removed from a fistula between the fundus of the gallbladder and the first part of the duodenum. Cholecystectomy was performed and the duodenal fistula was repaired. The cholecysto-colic fistula was repaired in two layers and a loop ileostomy was created to decompress the difficult duodenal closure after gastric resection. A T-tube duodenostomy in the management of duodenal stump was originally described by Dardik et al. in 1973.7

Discussion

In 1893 Leon Bouweret first described this syndrome, in which one or more gallstones pass into the duodenum through a biliary fistula causing gastric outlet obstruction. Bouweret’s syndrome complicated with a cholecysto-colic fistula is rare, with only one case reported in the literature; this patient died. The mortality rate of Bouweret syndrome can reach 30%. Rigler’s triad of pneumobilia, mechanical bowel obstruction and ectopic stone on plain abdominal radiograph can assist in making the diagnosis.4 The abdominal radiograph may also show a dilated stomach. An abdominal ultrasound scan may detect ectopic gallstones. However, a CT scan and endoscopic retrograde cholangiopancreatography (ERCP) remain the most sensitive modalities for diagnosis.

Although endoscopy is considered the first-line treatment, especially in high-risk patients, bowel obstruction due to migration of the fragmented stones may occur, necessitating urgent laparotomy.6 Laparoscopic trans-pyloric extraction of the stone has been used safely.4 However, laparotomy is indicated if the stones are large or impacted and cannot be dislodged endoscopically.

Tube duodenostomy is indicated when there is concern about increased intraluminal duodenal pressure or passage of gastro-intestinal contents through the duodenum after duodenal repair.6 Welch and Rodkey described tube duodenostomy to decompress a difficult duodenal closure after gastric resection.6 T-tube duodenostomy in the management of duodenal stump was originally described by Dardik et al. in 1973.7

Conclusion

Bouweret’s syndrome with cholecysto-colic fistula is a rare clinical entity associated with high mortality. Early recognition of the syndrome, creating a controlled duodenal fistula, early postoperative enteral feeding and enhanced postoperative care all helped achieve a favourable outcome in our patient.

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