Tension Pneumothorax, Unusual Presentation of Post-ERCP Duodenal Perforation. Case Report and Literature Review

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Abstract

INTRODUCTION
Since Meinhard Classen performed the first endoscopic retrograde cholangiopancreatography (ERCP) in 1973, this procedure is gaining more and more popularity in diagnosis and treatment of biliary and pancreatic diseases. ERCP is widely regarded as safe, but the major complication rate approaches 10%. Common complications include pancreatitis, bleeding, cholangitis, and perforation. ERCP-related perforations occur in about 1% of patients and this injury carries a death rate of 16% to 18%. Tension pneumothorax complicating ERCP has been reported but is a rare occurrence. We report a case of right side tension pneumothorax complicating an ERCP in a 65-year-old lady who had an ERCP for common bile duct stones. We also review the literature for the different options available for management of common bile duct stones and their risks.

CASE REPORT
A 65-year-old female with no co-morbidities presented to the emergency room of King Fahad Medical City, a tertiary care center in Riyadh, Saudi Arabia, with chief complaints of right upper quadrant pain and vomiting for 5 days. On examination, she was conscious, oriented, jaundiced and hemodynamically stable; she was afebrile and had an oxygen saturation of 94% on room air. She had mild right hypochondriac tenderness, no organomegaly, no palpable lump or free fluid on abdominal examination. Her other systemic examinations were unremarkable. Laboratory investigations revealed hemoglobin of 13g/dl with leukocyte count of 14.0 x 10³, predominantly neutrophilic (polymorphs 92%). The liver function tests showed total bilirubin of 21.6 umol/L, direct bilirubin of 9.7 umol/L, ALT 317 umol/L, AST 300 umol/L, alkaline phosphatase 488 umol/L, GGT 498 umol/L, and amylase 943 umol/L. Her renal function test and electrolytes were normal. Abdominal ultrasound revealed a prominent CBD at the porta hepatis, intrahepatic biliary ductal dilatation and cholelithiasis (figure 1 and 2).

Figure 1
Figure 1: Ultrasound showing a contracted gallbladder with gall stones.
Figure 2
Figure 2: Doppler ultrasound showing dilatation of the proximal part of the common bile duct anterior to the portal vein.

She was started on antibiotics and intravenous fluids. She had an ERCP which showed a duodenal diverticulum just across and proximal to the ampulla (figure 3).

Figure 3
Figure 3: Endoscopic view showing the duodenal diverticulum.

Cholangiogram revealed a dilated CBD with four filling defects suggestive of CBD stones (figure 4).

Figure 4
Figure 4: ERCP Cholangiogram showing a dilated CBD with filling defects suggestive of CBD stones

Due to difficulty in cannulation of the CBD, a precut sphincterotomy was performed with a needle knife and successful selective CBD cannulation was achieved.

Four stones were successfully removed from the common bile duct after extending the sphincterotomy with the sphincterotome. Later, balloon occlusion cholangiogram did not show any filling defect. Soon after the procedure, the patient became tachypneic, and hypoxic. The O2 saturation dropped to 66%, and she became hypotensive. Chest examination revealed deviation of trachea and mediastinum to the left with hyperresonant percussion note and absent breath sound on the right hemithorax. There was abdominal distension with diffuse mild tenderness. Chest x-ray confirmed right-sided pneumothorax, and free air under the right hemi-diaphragm (Fig.5).
A wide-pored cannula was placed in the 2nd intercostal space anteriorly, partially relieving her distress, but there was marginal improvement in her oxygen saturation. A chest tube was placed in the right side by the thoracic surgeon, and she was intubated and placed on mechanical ventilator. The patient’s oxygen saturation improved, but she remained hypotensive requiring inotropic support and broad spectrum antibiotics. Subsequently an exploratory laparotomy was performed, which revealed a 3cm tear over the lower anteriolateral aspect of the second part of the duodenum (Fig.6). No bile duct perforation was visualized. The duodenal laceration was sutured in 2 layers and reinforced by an omental patch. Cholecystectomy was performed at the same time.

The chest tube was removed after 5 days. Her hospital course was complicated with recurrent pneumonia and intra-abdominal fluid collection around the retroperitoneal part of the duodenum which was drained percutaneously under CT guidance with placement of a drainage catheter. The patient recovered from the surgery, and was later discharged in a stable condition. She was followed in the outpatient clinic for 2 years and she did not have evidence of duodenal stenosis.

**DISCUSSION**

Approximately 10% of patients with symptomatic gallstones may have associated common bile duct stones (1).

Whereas laparoscopic cholecystectomy represents the gold standard treatment for gallstones, there is no universal consensus on the optimal treatment of common bile duct stones (2,3). The predictive value of non-invasive tests as well as the preoperative diagnosis and management of common bile duct stones have not been well defined (4).

The treatment options available are various: preoperative endoscopic retrograde cholangiopancreatography with or without endoscopic biliary sphincterotomy, laparoscopic CBD exploration (transcystic or transcholedochal), or laparotomy with CBD exploration (by T-tube, C-tube insertion, or primary closure) (2, 5).
Traditional open choledochotomy remained the standard treatment of common bile duct stones for many years since Knowsley Thornton successful performed the first common bile duct exploration in 1889 (6), only 7 years after Carl Augustus Langenbuch performed the first cholecystectomy on July 15, 1882 at the Lazarus Hospital in Berlin (7).

The advent of laparoscopic cholecystectomy, first performed by Erich Mühe of Böblingen, Germany, in 1985 (8), has reopened the debate on the optimal management of patients with CBD stones or suspected CBD stones undergoing cholecystectomy.

Patients with common bile duct stones undergoing laparoscopic cholecystectomy may be managed by laparoscopic common bile duct exploration at the time of surgery, or undergo peri-operative ERCP. There is no evidence of a difference in efficacy, morbidity or mortality when these approaches are compared, though laparoscopic common bile duct exploration is associated with a shorter hospital stay (8). Numerous studies, including the European Association of Endoscopic Surgery (EAES) multicentre prospective randomized trial by Cuschieri et al., have shown equal efficacy in terms of ductal stone clearance for these two management options. (10, 11)

The two approaches are considered equally valid treatment options; however, laparoscopic common bile duct exploration is a tedious operation and requires both experience and facilities which are only available in advanced laparoscopic centers (12). Due to experience and technical availability, preoperative ERCP and laparoscopic cholecystectomy is most-often preferred.

For many surgeons, preoperative endoscopic sphincterotomy is the procedure of choice for managing common bile duct stones. The combination of endoscopic sphincterotomy and laparoscopic cholecystectomy provides a two-stage minimally invasive approach. This is surgically easy and saves operative time (13). Although early cholecystectomy is advised, there is no consensus about the time interval between laparoscopic cholecystectomy and ERCP.

Theoretically, as the interval between the ERCP and cholecystectomy increases, the rate of complications including acute cholecystitis, obstructive jaundice and acute pancreatitis increases as well.

Zafer compared 200 patients in whom ERCP was immediately followed by laparoscopic cholecystectomy with 200 patients who had interval cholecystectomy after ERCP. He concluded that ERCP followed by immediate laparoscopic cholecystectomy was safe with significant reduction in the average length of stay and average cost of treatment per patient as compared to ERCP followed by interval laparoscopic cholecystectomy. The complications were not significant in either group (14). Similarly Salman et al. concluded that early cholecystectomy after ERCP, within 72 hours, has better outcomes than delayed cholecystectomy (after 72 hours) (15).

Some authors suggest that a one-stage ERCP and laparoscopic cholecystectomy for the management of cholecysto-choledocholithiasis is safe and an effective technique with a low rate of post-ERCP pancreatitis. It offers another alternative for surgeons, especially those who do not practice laparoscopic common bile duct exploration to treat patients in a single setting (16, 3).

Since Meinhard Classen performed the first ERCP in 1973, this procedure is gaining more and more popularity in diagnosis and treatment of biliary and pancreatic diseases. Recently, it has been used in lithotripsy, endoprosthesis placement, stone extraction, and sphincterotomy (17).

ERCP is widely regarded as safe, but the major complication rate approaches 10%. Common complications include pancreatitis, bleeding, cholangitis, and perforation. Overall, the procedure carries a death rate of 1.0% to 1.5% (18, 19). ERCP-related perforations occur in about 1% of patients and this injury carries a death rate of 16% to 18% (20).

Little is known about the pathology of duodenal diverticula. The duodenum holds the second place in frequency of diverticula in the digestive tract after the colon. Duodenal diverticula are rare and they are asymptomatic in 90% of cases. Infrequently they can be a cause of upper gastrointestinal (GI) hemorrhage. Precise preoperative diagnosis is occasionally difficult (21). Diagnosis is usually made by endoscopy, contrast X-rays of the upper GI tract and as an intra-operative finding (22).

Periampullary diverticula discovered incidentally during endoscopic retrograde cholangiopancreatography are usually asymptomatic, but can be a source of significant morbidity (23). Cannulation of the common bile duct is more difficult in patients with periampullary diverticulum and requires more skills. Periampullary diverticulum is also associated with higher risk of retained stones in the common bile duct.
which may lead to higher post-endoscopic retrograde cholangiopancreatography complication rate (24).

Our patient had a large duodenal diverticulum across the ampulla on the lateral wall. Care was exercised not to hit the lateral duodenal wall during the process of stone extraction. As the ampullary cannulation was difficult, a decision for pre-cut needle knife sphincterotomy was made. The long duration of ERCP with accompanied insufflation of air for lumen patency can predispose to rupture of duodenal diverticulum, with subsequent pneumoperitoneum and tension pneumothorax. This is the most plausible hypothesis explaining tension pneumothorax in our patient, as at the time of exploration no other injury site including the CBD was identified.

ERCP-related perforations can present as retroperitoneal duodenal perforations which are most common, as free bowel perforations, or perforations of bile duct.

Stapfer et al. (25) classified the perforations in relation to mechanism, anatomical location, and severity of injury, which may predict the need for surgery. They classified ERCP-related perforations into 4 types in descending order of severity: Type I, lateral or medial wall duodenal perforation; Type II, peri-Vaterian injuries; Type III, distal bile duct injuries related to wire/basket instrumentation; Type IV, retroperitoneal air alone. Type IV is probably related to the use compressed air to maintain the patency of the duodenal lumen, which can result in air diffusion within the layers of duodenal lumen wall or outside the lumen, as in pneumatosis cystoides (25-27).

Pneumothorax is a serious but rare complication of ERCP [28-31]. It can be a result of both intraperitoneal duodenal perforation [30] and retroperitoneal duodenal perforation (29).

There are various clinical courses and presentations of post-ERCP duodenal perforations depending on the extent of the perforation. However, the development of pneumothorax in patients undergoing ERCP is rare. Lee et al. reported tension pneumothorax with a complicating micropereforation following ERCP (32). Similarly, Hui et al. reported a case of tension pneumothorax complicating ERCP in a patient with Billroth II gastrectomy (28).

Macklin and Macklin described the phenomenon of extralveolar air leak from high intrapulmonary airway pressures (33, 34). Air dissents along perivascular sheaths to the mediastinum and thence to the neck, subcutaneous tissues, and extraperitoneal spaces. Unabated airway pressure can force air from the retroperitoneum into the free peritoneal cavity with resulting pneumoperitoneum.

Some authors claimed that the process is reversed in cases of pneumothorax complicating post-ERCP duodenal perforation as continued air insufflation into the retroperitoneal space ultimately leads to pneumomediastinum and pneumothorax. Alternatively, the pneumothorax could be explained on the basis of communications between the pleural space and the peritoneum by pleuroperitoneal defects [33, 35, and 36].

Diaphragmatic pores or “stomata of von Recklinghausen” form when the cell margins of lymphatic endothelial and mesothelial cells lining the peritoneal surface of the diaphragm come together, which provide an open channel between the peritoneal cavity and lymphatic lacunae. There are a variety of so-called porous diaphragm syndromes in which fluids (e.g., acites), gases (from a therapeutic or spontaneous pneumoperitoneum), tissue (endometrium with catamenial pneumothorax or hemothorax), secretions (from a subhepatic abscess, pancreatic pseudocyst, bilothorax), and intestinal content (perforated peptic ulcer) may pass. A large bore is not needed for tension pneumothorax. It can develop even one or two days later if intestinal air leakage is continued (18). These pathways are invoked to explain tension pneumothorax associated with tension pneumoperitoneum.

Early recognition and appropriate management are essential to optimal output in perforations complicating ERCP. Surgical indications after duodenal perforation are acute peritoneal irritation signs with or without sepsis, documentation of large contrast extravasation, presence of intra- or retroperitoneal fluid collections, or suspected perforation with retained material. In the absence of these indications and if the perforation is recognized at an early stage, conservative management can be attempted (37). According to Stapfer et al., type 1 perforations were usually managed by surgery, while surgery is less likely in other types (27).

**CONCLUSION**

Whereas laparoscopic cholecystectomy represents the gold standard treatment for gallstones, there is no universal consensus on the optimal treatment of common bile duct stones. The advent of laparoscopic cholecystectomy has
reopened the debate on the optimal management of patients with common bile duct stones or suspected common bile duct stones undergoing cholecystectomy.

ERCP is widely regarded as a safe procedure, but the major complication rate approaches 10%. Common complications include pancreatitis, bleeding, cholangitis, and perforation. Overall, the procedure carries a death rate of 1.0% to 1.5%. ERCP-related perforations occur in about 1% of patients and the injury carries a death rate of 16% to 18%.

Periampullary diverticula make cannulation of the common bile duct more difficult and may predispose to ERCP-related duodenal perforations.

Tension pneumothorax is a rare life-threatening complication of ERCP duodenal perforation which requires immediate recognition and appropriate management.

References

29. Gya D, Sali A, Angus D: Subcutaneous emphysema and
34. Macklin MT, Macklin CC: Malignant interstitial emphysema of the lungs and mediastinum as an important occult complication in many respiratory diseases and other conditions: an interpretation of the clinical literature in light of laboratory experiments. Medicine; 1944; 23: 281-358.
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