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BRIEF REPORT

Post-endoscopic retrograde cholangiopancreatography pneumothorax: Report of two cases and literature review

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Introduction

Endoscopic retrograde cholangiopancreatography (ERCP) is a widely used therapeutic modality in the management of pancreaticobiliary diseases. The incidence of major complications ranges from 5.4 to 23%. Pancreatitis, bleeding, perforation, and cholangitis are relatively common complications of ERCP. The overall mortality rate is <1%. However, delayed diagnosis and intervention of duodenal perforation carries a very high mortality rate (8–23%).[1] Pneumothorax is a very rare complication of ERCP, with only a handful of cases described in the literature.[2–17] Here, we describe two cases of post-ERCP pneumothorax and systematic literature review.

Case report

Case-1. A 65-year-old hypertensive female presented with abdominal pain. Blood investigation showed serum alkaline phosphatase of 267 U/L (normal 43–115 U/L); otherwise, tests were normal. Ultrasonography demonstrated a bile duct stone sized 6 mm. ERCP was performed under general anesthesia. Bile duct access was achieved using a wire-guided cannulation technique. A filling defect of <1 cm size was noted in the lower bile duct. The patient developed unexpected oxygen desaturation immediately after the sphicterotomy. She was noted to develop subcutaneous emphysema (SCE) extending from the upper thorax up to the eyelids. Decreased air entry and hyperresonant notes were present on the right side of chest. The procedure was abandoned, and the patient was immediately transferred to the intensive care unit (ICU). A chest x-ray showed right side pneumothorax. Computed tomography (CT) scan demonstrated right pneumothorax, pneumomediastinum, and pneumoretroperitoneum (Fig. 1a,b). There was complete collapse of the middle lobe and partial collapse of basilar segments of the lower lobe of the right lung. Air bubbles were seen in the subcutaneous tissue of the anterior chest wall. There was no leakage of intestinal contents into the peritoneal cavity (Stapfer type IV perforation). The patient did not develop symptoms of peritonitis. Patient was kept nil per oral, and a nasogastric tube was placed. The conservative treatment was performed with antibiotics, intravenous fluids, chest tube drainage and mechanical ventilation. Two days later, SCE started decreasing, pneumothorax regressed, and the patient was extubated. One week later, the patient was discharged in healthy condition. ERCP and stone extraction was performed 4 weeks after discharge.

Case-2. A 25-year-old woman with cholelithiasis and choledocholithiasis was referred for ERCP. Cholangiogram demonstrated a stone of about 1.2 cm in size in the bile duct. Sphincterotomy

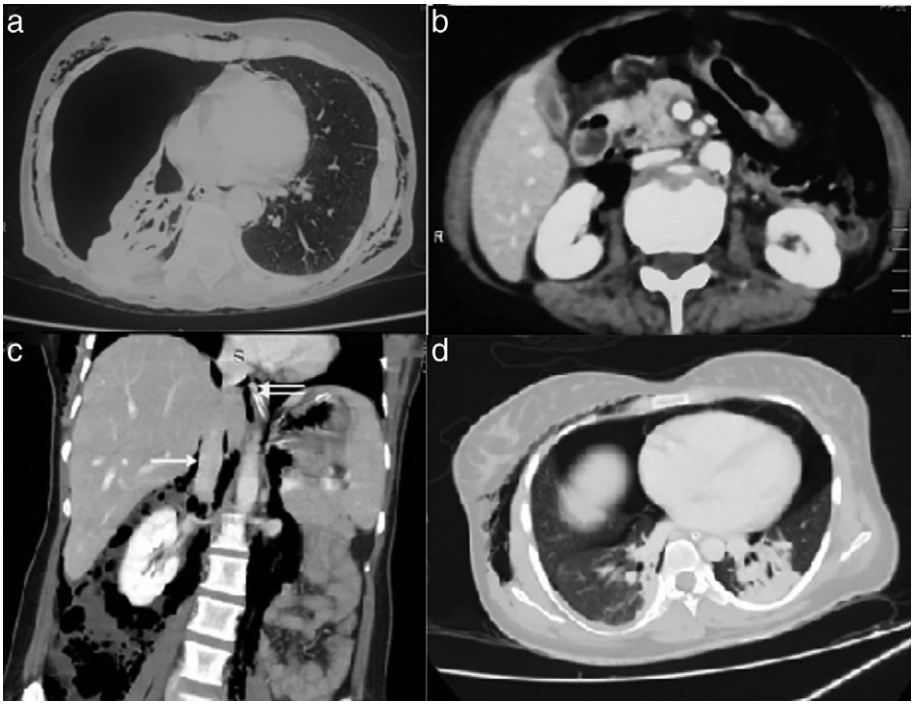

Figure 1 Computed tomography (CT) scan shows (a) right pneumothorax with passive collapse of basilar segments of lower lobe of right lung; subcutaneous emphysema present in the bilateral anterior chest wall, (b) gas accumulation in the right perinephric space, and (c) extensive gas accumulation in the perinephric spaces (right > left). Note the cranial extension of the air along the inferior vena cava (IVC) and aorta (arrow) into the thoracic cavity, (d) bilateral pneumothorax, and subcutaneous emphysema.

was performed. The patient developed oxygen desaturation, abdominal distension, and SCE soon after the sphicterotomy. The procedure was abandoned, and the patient was shifted to the ICU. CT scan showed bilateral pneumothorax, pneumomediastinum, SCE, pneumoperitoneum, and pneumoretroperitoneum (Fig. 1c,d). The patient developed signs of peritonitis. Laprotomy and repair of a rent (2 cm) located in the posterolateral wall of the second portion of duodenum (Stapfer type 1 perforation) was performed. The patient was subsequently discharged in healthy condition.

Discussion

We report two cases of pneumothorax following ERCP for choledocholithiasis that were diagnosed over a 4-year period in a teaching hospital. Unexplained oxygen desaturation during or after ERCP procedure with and without the presence of an abdominal sign of perforation warrant further investigation for possible air leak syndrome. Early diagnosis and prompt management is essential to improve the outcome in such cases.

ERCP-related perforations are classified by Stapfer *et al.* into four types based on severity and anatomical location (type I: lateral or medial duodenal wall perforations; type II: peri-Vaterian injury; type III: bile or pancreatic duct injury; and type IV: presence of retroperitoneal air alone).[18] Early diagnosis and prompt treatment decisions are essential to avoid mortality in patients due to perforation. Treatment of ERCP-related perforations depends on the type of injury and clinical condition of patient. Surgical indications after duodenal perforation are peritonitis with or without sepsis, documentation of large contrast extravasations, presence of intra- or retroperitoneal fluid collections, or suspected perforation with retained material. Type I perforations require immediate surgery. Most of type II perforations tend to seal spontaneously within 48–72 h, but nearly one-third of patients may require surgical repair. Type III perforations usually close spontaneously and can be conservatively managed with stent placement. Type IV perforations may not have an overt transmural perforation, mostly treated conservatively. We did not find an overt transmural perforation in index case 1 and managed the case successfully with conservative management.

The proposed mechanisms of post-ERCP pneumothorax and SCE are as follows (Fig. 2): (i) Air enters the retroperitoneal space after interruption of the duodenal barrier, tracking from the retroperitoneal space to the peritoneum, pleural space, mediastinum, and subcutaneous tissue. The spread of air takes place possibly through deep fascial planes.[2,19] The deep fascia in the neck surrounds the trachea and esophagus and is contiguous with the diaphragmatic and/or esophageal hiatus and major airways of the thorax. (ii) After perforation, the air flows from the duodenum to the right anterior pararenal space, then to the posterior pararenal space from where the air can reach the diaphragmatic hiatus, causing pneumomediastinum, pneumothorax, or cervical SCE. This is supported by the fact that the CT scan in the present case 1 showed retroperitoneal gas in perinephric space without the presence of pneumoperitoneum. (iii) Another hypothesis is that air spread along the perineural and perivascular sheaths to enter the mediastinum. (iv) Two less possible pathways are through porous diaphragm or alveolar rupture due to valsalva manoeuvre.[20]

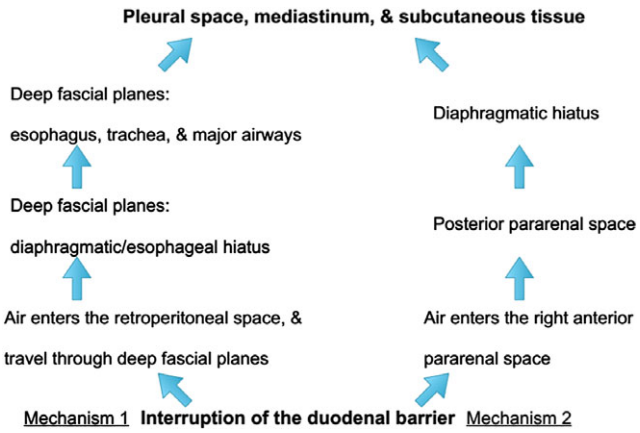

Figure 2 Flow diagram showing the major mechanisms of post-endoscopic retrograde cholangiopancreatography air leak syndrome.

Earlier reports suggest that a complete transmural defect is not essential for tracking of air from the duodenal lumen to retroperitoneum. This is supported by the fact that approximately one-third of reported cases of pneumothorax are unrelated to sphincterotomy (Table 1). Local weakening of the duodenal–retroperitoneal barrier secondary to duodenal diseases may cause leakage of air into the retroperitoneum.

Post-ERCP pneumothorax is a rare but potentially under-reported complication. To the best of our knowledge, a review of the English literature showed 35 cases of post-ERCP pneumothorax. The salient features of 37 cases (including the present cases) are shown in Table 1. Pneumothorax is usually bilateral (51.35%) and right sided (37.83%) and is mostly accompanied by pneumomediastinum and SCE. Retroperitoneal and intraperitoneal air can be absent in nearly one-fourth of cases. Clinical features of peritonitis can be absent in these patients and may cause a diagnostic dilemma. The potential risk factors, such as sphincterotomy, juxtapappilary divercula, female gender, and older age (>60 years), are present in 67.56, 16.21, 78.37, and 62.16% of patients, respectively. The patients in the present case series were female and underwent sphincterotomy. In our opinion, the endoscopist's experience, the proper techniques of sphincterotomy, and prolonged procedure time are the other important risk factors of air leak syndrome. The endoscopist must be very careful while performing sphincterotomy, especially in elderly female patients. Most cases are successfully treated conservatively with nil orally, nasogastric tube placement, antibiotics, and chest tube drainage when indicated. Surgery is required in nearly one-fourth of cases. Survival is noted in more than 90% of patients of ERCP complicated by pneumothorax.

Table 1 Post-ERCP pneumothorax: Review of literature

Total cases	37
Mean (range) age	65.27 (23–90) years
Age > 60 years	23/37 (62.16%)
Gender (male:female ratio)	8:29
Indication of ERCP	Bile duct stone 22(59.45%); others 15 (40.54%)
Sphincterotomy done	25 (67.56%)
Juxta papillary diverticula	6 (16.21%)
Pneumothorax location	Bilateral 19 (51.35%); right side 14 (37.83%); left side 4 (10.81%)
Retroperitoneal air absent	7 (18.91%)
Intraperitoneal air absent	11 (29.72%)
Treatment	Conservative (±chest tube) 26 (70.27%); surgery 11 (29.72%)
Mortality	3 (8.10%)

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