Massive Subcutaneous Emphysema following Endoscopic Retrograde Cholangiopancreatography with Sphincterotomy

Michail Papamichail a  Nicolas Nikolaidis b
Elina Anastasiou c  George Sidirokastritis c
Panagiotis Prigouris c

a Northwick Park & St Mark’s Hospital, London, and b Southampton University Hospital, Southampton, UK; c Evangelimos General Hospital of Athens, Athens, Greece

Key Words
Endoscopic retrograde cholangiopancreatography · Pneumomediastinum · Pneumoperitoneum · Subcutaneous emphysema

Abstract
Although endoscopic retrograde cholangiopancreatography (ERCP) is an effective procedure for the diagnosis and treatment of the pancreatic and extrahepatic biliary tract diseases, it is still related with several complications. A female patient who underwent an ERCP with sphincterotomy developed massive subcutaneous emphysema along with pneumomediastinum and pneumoperitoneum. Although mild respiratory distress occurred, based on the absence of intraabdominal leakage of gastrografin, the patient was managed conservatively. In conclusion, the retroperitoneal air collection related to ERCP is well recognized even in the absence of obvious perforation and may spread to adjacent areas, causing serious complications.

Introduction
Endoscopic retrograde cholangiopancreatography (ERCP) is a well-established effective procedure for the diagnosis and treatment of pancreatic and extrahepatic biliary tract diseases [1]. There is general consensus that ERCP should be performed for specific indications, by trained endoscopists, using standard techniques with careful monitoring of the patient during and after the procedure [2, 3]. Although nowadays it is routinely...
used and is considered to be a safe procedure, especially in expert hands [1], it still carries significant morbidity [4, 5]. Even in most experienced centres common complications such as pancreatitis (3.2%), duodenal or common bile duct perforation (0.12%), haemorrhage (0.62%), infection (0.75%) and general cardiac and pulmonary complications (0.25%) may occur [6]. In addition a group of miscellaneous complications (1.3%) is also well recognized [7]. All of the above may well be related to operator experience, method variations and individual patient characteristics [8, 9].

One of the most feared, unpleasant sequelae after ERCP is air leakage, which originates either from the duodenum and small bowel or from the extrahepatic bile duct system. In our institution such an unusual event occurred regarding a female patient who developed post-ERCP massive subcutaneous emphysema along with pneumomediastinum and pneumoperitoneum.

**Case Report**

An 83-year-old woman presented with symptomatic cholelithiasis. Initially, due to her old age and significant co-morbidities (left-sided heart failure, atrial fibrillation and diabetes), she was managed conservatively. On the third day after admission, she became febrile (38.1°C) with leukocytosis of 13,300/mm³ (normal 4,100–10,900/mm³) accompanied with rigor, increased abdominal tenderness and slight jaundice. Bilirubin (5.1 mg/dl; normal 0–1.5 mg/dl) and alkaline phosphatase (550 mg/dl; normal 25–130 mg/dl) were elevated, but due to low serum amylase (115 mg/dl; normal 20–130 mg/dl) and urine amylase (480 mg/dl; normal 0–650 mg/dl), acute pancreatitis was excluded. The patient underwent an ERCP with sphincterotomy and extraction of choledochal sludge suggesting a diagnosis of acute cholangitis. Over the following 24 h she developed massive subcutaneous emphysema with extensive swelling and palpable crepitations over the trunk, neck, face and upper extremities. She underwent an abdominal and thoracic computed tomography (CT) scan which revealed significant pneumomediastinum and pneumoperitoneum, but there was no intraabdominal leak of gastrografin (fig. 1, fig. 2, fig. 3, fig. 4). The patient had mild symptoms of respiratory distress with slight tachypnea (25–30 pm) and free air oxygen saturation 95%. Furthermore no electrocardiographic abnormalities (except for atrial fibrillation) or other signs of cardiovascular compromise (blood pressure 130/70 mm Hg, heart rate 110 bpm) were detected. Since there was no radiological evidence of perforation and no clinical sign of subsequent peritonitis, she was managed conservatively with close monitoring of the respiratory and cardiovascular status. The patient responded well and gradual resolution of the subcutaneous emphysema occurred within 4 days.

**Discussion**

The presence of subcutaneous emphysema after an ERCP is a rare but well-recognized complication [5, 10, 11]. The most common cause is duodenal perforation which results in retroperitoneal collection of free air [4, 5, 12]. However even in the absence of an obvious perforation ERCP is well correlated with the evidence of retroperitoneal air, which is not an uncommon finding on CT scans when performed after ERCP [1]. This probably occurs due to the insufflation of pressurised air in order to maintain the patency of the lumen (gastrointestinal or biliary tract) in combination with co-existing mucosal disruption or a small iatrogenic perforation not detected by imaging techniques [1, 5, 11, 13]. This air may spread through the facial planes to the subcutaneous tissue, leading to subcutaneous emphysema, or diffuse to the peritoneal cavity, causing pneumoperitoneum, or even escape through tiny communications between the pleural space and the peritoneum, causing pneumothorax or pneumomediastinum [4, 5, 11, 13]. An alternative pathway in creating pulmonary interstitial emphysema is alveolar rupture
due to increased intrathoracic pressure secondary to vomiting following ERCP. In this case air reaches the mediastinum through the adjacent bronchovascular pleural sheaths [13]. In case of significant amount of air all the above may occur like in our case with massive subcutaneous emphysema.

In conclusion, retroperitoneal air collection related to ERCP is well recognized even in the absence of obvious perforation. Its treatment is basically non-surgical unless a gastrointestinal or biliary tract perforation has been established.

**Competing Interests**

The authors declare that they have no competing interests.

**Authors’ Contributions**

M. Papamichail analyzed and interpreted the patient data regarding the initial diagnosis and the subsequent complication and was a major contributor in writing the manuscript. N. Nikolaidis and E. Anastasiou were major contributors in writing the manuscript. G. Sidirokastritis performed the ERCP. P. Prigouris co-ordinated the management of the patient and of the authors’ writing. All authors read and approved the final manuscript.

**Fig. 1.** CT scan of the chest. Axial view at the level of the aortic arch showing extensive surgical emphysema spreading anteroposteriorly in the subcutaneous tissues. There is also widespread pneumomediastinum around the trachea and the great vessels.
**Fig. 2.** Lung window.

**Fig. 3.** CT scan of the abdomen following oral contrast (gastrografin). Axial view at the level of the transverse colon demonstrating air embracing the small bowel loops and infiltrating the omentum. There is no obvious gastrografin leak.
Fig. 4. Lung window.

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Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review from the journal’s Editor-in-Chief.