

CASE REPORT

Massive subcutaneous emphysema, unilateral pneumothorax, pneumomediastinum and pneumoperitoneum after endoscopic retrograde cholangiopancreatography

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Abstract

In this paper we discuss a case of duodenal perforation resulting in subcutaneous emphysema, left-sided pneumothorax, pneumomediastinum and pneumoperitoneum. Duodenal perforation is a rare, but serious complication of endoscopic retrograde cholangiopancreatography (ERCP). Signs indicating perforation include agitation, tachypnoea, dyspnoea, subcutaneous emphysema or hypoxaemia. Diagnosis can be confirmed by abdominal computed tomography (CT) scan. Treatment is surgical or conservative and based on the type of perforation, clinical status and radiographic imaging. Conservative treatment consists of the administration of oxygen, broad spectrum antibiotic therapy, biliary and duodenal drainage (nasobiliary and/or nasogastric tubes), nil by mouth, and if indicated, with respect to the severity of pneumothorax and hypoxaemia, uni- or bilateral pleural drainage. Frequent re-evaluation of the patient's clinical condition is warranted.

Introduction

Endoscopic retrograde cholangiopancreatography (ERCP) is an endoscopic procedure used for the diagnosis and management of biliary and pancreatic disorders. Complications include bleeding, perforation and pancreatitis. Duodenal perforation is a rare, but dangerous complication and will be discussed in this case report.

Case

An 89-year-old female patient underwent an elective endoscopic retrograde cholangiopancreatography (ERCP) because of persistent choledocholithiasis after recent cholecystectomy. During ERCP the patient was in a prone position. The procedure was technically challenging, because the ampulla was located in a large duodenal diverticulum.

During the procedure, before sphincterotomy, the patient became restless upon which the procedure was terminated and she was positioned on to her back. After a short while her respiratory distress worsened and she spontaneously developed substantial subcutaneous emphysema. Because of the respiratory distress and desaturation the patient was transferred to the Intensive Care Unit and immediately intubated. A chest X-ray revealed subcutaneous emphysema (*figure 1*). A thoracic and abdominal computed tomography (CT) scan showed a left-sided pneumothorax, pneumopericardium, pneumomediastinum, pneumoperitoneum and extensive subcutaneous emphysema (*figure 2*). It also showed

Figure 1. Chest X-ray showing extensive subcutaneous emphysema



Figure 2. CT image showing left sided pneumothorax, pneumomediastinum, pneumopericardium and subcutaneous emphysema



Figure 3. Patient after intubation. Subcutaneous emphysema of the chest, neck, shoulders and face



a duodenal diverticulum with two pockets of contrast outside of the duodenum. This finding was considered suspect for extravasation of contrast in the free abdominal space. Because of hypoxaemia, a chest tube was inserted to resolve the left-sided pneumothorax. In view of the patient's clinical stability and her advanced age, associated with a high risk during surgical interventions, it was decided not to push for surgery immediately. The patient was given a nasogastric tube for gastric decompression, intravenous broad spectrum antibiotics and total parenteral feeding was started (*figure 3*). There was no contained retroperitoneal fluid collection amenable to drainage. On day two the chest tube was removed and patient could be weaned from mechanical ventilation. Subcutaneous emphysema resolved within a few days and the patient was extubated. On day seven oral intake was restarted. No further complications occurred and patient was discharged from the ICU. She was discharged from hospital and is currently in good health.

Discussion

ERCP was introduced in 1968¹ and since then it has developed into the preferred therapy for many biliary and pancreatic disorders like choledocholithiasis, for the diagnosis and treatment of biliary and pancreatic neoplasms and for the postoperative management of biliary perioperative complications.² It is the treatment of choice for biliary duct stones. It is a technically challenging procedure with potentially serious complications. The most commonly occurring complications are pancreatitis, bleeding, sepsis and perforation. The overall complication rate is 4-10%.

Duodenal perforation has the lowest incidence level, ranging from 0.1% to 0.6%.³ Perforation is more often retroperitoneal which has the highest occurrence rate^{3,4} and usually occurs after papillotomy. Other types of duodenal perforation are free bowel wall perforation and perforation of the bile duct. Duodenal perforation can lead to retroperitoneal collection of free air resulting in subcutaneous emphysema. Subcutaneous emphysema after ERCP was first reported in 1989.⁵ The mortality rate in patients with perforation can be high (16-18%) if there is delayed diagnosis or treatment.⁶ However, more recent studies have shown a lower mortality rate of 8%.⁷ Deaths are related to sepsis and peritonitis as a consequence of duodenal perforation.

Clinical signs that could indicate a perforation, especially after sphincterotomy, are agitation, tachypnoea, dyspnoea, subcutaneous emphysema or hypoxaemia. For this reason it is vital to monitor oxygen saturation during the procedure as recommended in several guidelines.⁸ The diagnosis of perforation can be confirmed by abdominal CT scan. A perforation is not always seen on routine thoracic radiographic images and the amount of retroperitoneal air does not correlate with the severity of disease.⁹

Risk factors for perforation include the performance of a sphincterotomy, Billroth II anatomy, the intramural injection of contrast, prolonged duration of procedure, biliary stricture dilation, and sphincter of Oddi dysfunction. A more recent

study found sphincterotomy and malignancy as additional risk factors for perforation.²

There is an ongoing debate whether a juxtapapillary diverticulum is a risk factor for duodenal perforation during ERCP. A recent prospective study showed no significant difference in perforation rate in patients with or without a duodenal diverticulum.¹⁰ Older studies, however, did report a juxtapapillary diverticulum as a risk factor for duodenal perforation. The prevalence of periampullary diverticula ranges from 9% to 32.8% in populations undergoing ERCP whereby in 6.8% to 54.9% of cases the papilla is located in a diverticulum.¹⁰ It is well known to endoscopists that gaining access to a papilla that is located within a duodenal diverticulum is challenging and therefore makes sphincterotomy more difficult to perform. Decisions on whether or not to operate on a patient are based on the type of perforation, clinical status and radiographic imaging. Perforations further away from the papilla usually require surgery, whereas the other types of perforation can be treated conservatively.¹¹ Conservative treatment consists of oxygen administration, broad spectrum antibiotic therapy, biliary and duodenal drainage (nasobiliary and/or nasogastric tubes), nil per mouth and frequent re-evaluation. If indicated, with respect to the severity of pneumothorax and hypoxemia, uni- or bilateral pleural drainage is needed. In up to 86% of cases, conservative treatment gives clinical resolution without requiring operative intervention.² Massive subcutaneous emphysema, however, is usually an indication for surgery.^{6,7} Advanced age contributes to poorer outcome in surgical treatment of ERCP perforations.¹²

A pneumothorax can be the result of retroperitoneal perforation. Usually a bilateral pneumothorax is found, but unilateral pneumothorax has been described before, mostly right-sided.⁴ The mechanism behind this type of pneumothorax remains unclear, but the leading hypothesis is that it is caused by insufflation of pressurised air via the iatrogenic perforation. This gives rise to spreading of air through the facial planes to the subcutaneous tissue, causing subcutaneous emphysema, and even between pleural space and peritoneum resulting in pneumothorax and pneumomediastinum.¹³ Another hypothesis is that air in the retroperitoneal space is pushed to the thoracic space by the diaphragmatic hiatus^{14,15} resulting in rupture of the parietal pleura⁴ and thus causing pneumothorax. Our patient developed respiratory insufficiency due to a combination of left-sided pneumothorax and a rigid chest wall due to excessive subcutaneous emphysema. No obvious upper airway compromise (notably no swelling of the larynx and pharynx) was seen during intubation. Our hypothesis is that insufflation of air during ERCP lead to rupture of the periampullary diverticulum that resulted in a massive air leak into the retroperitoneal space, followed by pneumomediastinum, pneumothorax and subcutaneous emphysema.

The amount of air (retroperitoneal and subcutaneous) did not correlate with the size of the perforation.

Conclusion

In short, our patient presented with massive subcutaneous emphysema, left-sided pneumothorax, pneumoperitoneum and pneumomediastinum due to perforation of a duodenal diverticulum during ERCP. This is a rare, but serious complication following ERCP. Treatment is generally conservative.

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