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Case Report

Concurrent acute pancreatitis, pneumoperitoneum, pneumoretroperitoneum, and pneumomediastinum following ERCP-related perforation: A rare and insightful case study ☆☆☆

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ABSTRACT

This case report details an extraordinary occurrence following endoscopic retrograde cholangiopancreatography (ERCP) in a 42-year-old woman. Despite ERCP being a commonly performed procedure, this case presented an unusual combination of acute pancreatitis, pneumoperitoneum, pneumoretroperitoneum, and pneumomediastinum resulting from a Stapfer type III perforation. The patient managed conservatively with nil per os, nasogastric tube, intravenous fluids, pain relief, and antibiotics, exhibited clinical improvement. Remarkably, resolution of complications occurred without surgical intervention. This case underscores the significance of vigilance in diagnosing and appropriately managing ERCP-related complications, contributing to the broader understanding of these rare events and fostering improved patient outcomes.

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Introduction

Endoscopic retrograde cholangiopancreatography (ERCP) is a routinely performed procedure for diagnosing and manag-

ing pancreaticobiliary disorders. Indications of ERCP include cholangitis, choledocholithiasis-associated biliary leak, acute pancreatitis, cholangiography, and biliary stent replacement or removal in benign or malignant diseases [1]. Although considered safe and impactful, the rate of complications after

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ERCP is 10%-12%. Frequent complications include pancreatitis (1.6%-15%), bleeding (0.3%-2%), cholangitis (0.5%-3%), cholecystitis (0.5%), and viscus perforation (1%) [2].

Stapfer has classified perforations into 4 types, usually identified through abdominal radiography and a computerized tomography (CT) scan of the abdomen in select cases [3]. Our case report is unique in that it describes the diagnosis and management of a patient with a *recherché* occurrence of acute pancreatitis with pneumoperitoneum, pneumoretroperitoneum, and pneumomediastinum developed simultaneously after an ERCP-related perforation.

Case report

A 42-year-old with 4 previous pregnancies sought medical attention at the gastroenterology department of the Pakistan Kidney & Liver Institute (PKLI) Hospital. She presented with epigastric pain for the past 2 years that worsened and radiated to her back for the past 2 months. The patient was normotensive and normoglycemic with no significant medical or surgical history. Liver function tests (LFTs) evinced raised total bilirubin (3.7 mg/dL; N: 0.1-1.0 mg/dL), ALT (650 IU/L; N: 10-40 IU/L), AST (720 IU/L; N: 12-38 IU/L), ALP (970 IU/L; N: 25-100 IU/L), and GGT (350 IU/L; N: 5-40 IU/L). Serum amylase and lipase levels were within normal limits. The patient underwent an ultrasonography scan, which revealed the presence of cholelithiasis and choledocholithiasis.

Consequently, an ERCP procedure was carried out. Following selective biliary cannulation and sphincterotomy, a cholangiography was performed, which demonstrated a single, partially impacted stone in the distal common bile duct (CBD) with a narrow caliber distal bile duct (Fig. 1). A small amount of sludge was removed with a balloon trawl; however, the stone could not be removed. Sphincterotomy was performed with a 10 mm controlled radial expansion balloon, and multiple attempts were made to retrieve the stone, but to no avail. Finally, a 7 Frx7 cm stent was placed for drainage. After stabilization, the patient was discharged.

One day post-ERCP, the patient presented to the emergency department with abdominal pain and relative constipation. On examination, the patient had tachycardia with a soft but tender abdomen, predominantly in the epigastrium and right hypochondrium, along with sluggish bowel sounds. Post-ERCP pancreatitis was suspected, for which the patient was readmitted. A CT scan of the patient was performed, which revealed extensive pneumoretroperitoneum, mild pneumoperitoneum, and pneumomediastinum (Figs. 2A–C). No collection or free fluid in the abdomen likely suggested ERCP-related sealed distal CBD/duodenal perforation. Changes of acute pancreatitis were also seen with a modified CT severity index of 4, along with cholelithiasis and a 10 mm stone in the distal CBD. The patient's white blood cell count, serum amylase, and serum lipase levels were also elevated.

The medical team conservatively managed the patient by administering nil per os (NPO), inserting a nasogastric tube, administering intravenous fluids, providing pain relief through intravenous medication, and administering

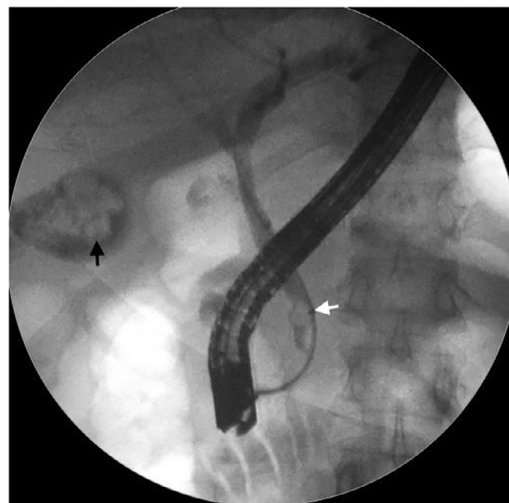


Fig. 1 – ERCP image illustrating a filling defect in the distal common bile duct (white arrow), indicative of calculus with subsequent narrowing of the duct. Additionally, multiple gallbladder calculi are evident in the image (black arrow).

broad-spectrum antibiotics. Following close observation for 5 days, the patient improved clinically with improvement in laboratory findings. A CT with oral contrast was performed that did not show any contrast extravasation, and an improvement was noticed in other results as well. The hospital staff subsequently discharged the patient.

An axial CT scan 6 weeks later demonstrated resolution of the pneumoperitoneum and pneumoretroperitoneum with unchanged cholelithiasis and a biliary stent in place (Fig. 3). The patient underwent a follow-up cholecystectomy, transduodenal stone extraction, and Roux-en-Y hepaticojejunostomy. Ten days after surgery, the patient presented with mild pus discharge from the wound site, managed by a daily dressing change. An ultrasonography scan of the patient revealed minimal collection at the porta hepatis. Subsequently, a follow-up after 2 weeks evinced a complete resolution of the collection.

Discussion

ERCP is a commonly used diagnostic and therapeutic technique for biliary and pancreatic disorders. Despite being a relatively safe procedure, it carries a higher risk of complications than other endoscopic procedures. Advancements in operator skills, endoscopic devices, and safety parameters have not decreased the incidence of ERCP-related complications [2]. Post-ERCP pancreatitis (PEP) is the most commonly encountered complication, with an average incidence of 3%-5% [2]. A systematic review by Kochar et al. [4] reported an overall PEP incidence of 9.7% with a mortality rate of 0.7%. The incidence varies primarily due to the changing definitions of PEP and its classification [2]. According to the current literature, there are 2 commonly used definitions of PEP: the Cotton definition [5] and the revised Atlanta International Consensus [6]. The

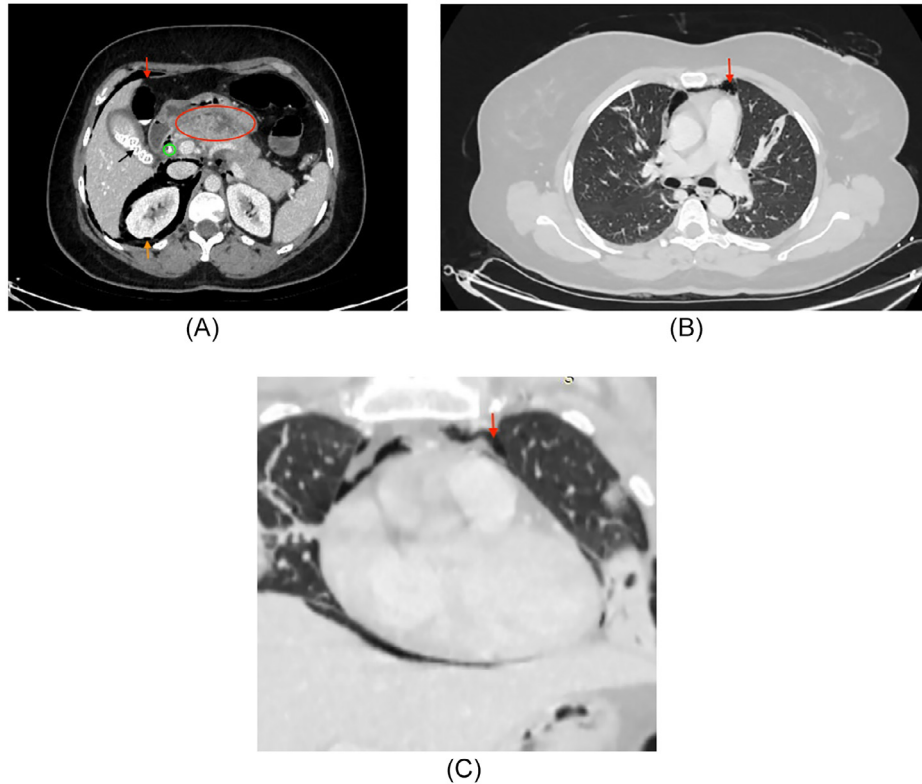


Fig. 2 – (A) Pneumoperitoneum, pneumoretroperitoneum, and acute pancreatitis: Axial CT at the level of the gallbladder reveals pneumoretroperitoneum (orange arrow) and pneumoperitoneum (red arrow), evident by free air in the retroperitoneum and peritoneum. Multiple calculi are observed within the gallbladder (black arrow), accompanied by mild diffuse edematous gallbladder wall thickening and the presence of an in-place biliary stent (green circle). The pancreas (red circle) displays mild edema with a focal area of reduced enhancement/necrosis. Minimal peripancreatic fat stranding is also discernible. (B and C) axial CT scan through the chest revealing mediastinal air (red arrow).

pathophysiology of PEP is not entirely understood, but it is believed to be caused by direct trauma during the procedure to the pancreatic duct, chemical insult from the injected contrast, infection due to the ascent of intestinal flora, or thermal injury from electrocautery [2].

On the other hand, although intestinal perforation is relatively rare (1%), it has a high mortality rate of 8%-23% [2]. The most common signs and symptoms of intestinal perforation include severe abdominal pain, tachycardia, leukocytosis, and fever [2]. Direct visualization with an endoscope, free air, or extravasated contrast on a CT scan helps diagnose duodenal perforation. In our case, a CT scan helped reach a presumptive diagnosis.

Howard et al. conducted a retrospective analysis of 6040 cases, out of which 48% underwent sphincterotomy, and found that 0.6% of patients developed perforation [7]. The study also reported an ERCP complication rate of 8.2% and a mortality rate of 1.3%. Duodenal perforation is a common complication that can be misdiagnosed as acute pancreatitis, as both can cause abdominal pain and vomiting [8]. To differentiate between these complications, performing serum amylase and imaging, specifically CT, is essential [8]. In our case, the patient's serum amylase levels were correlated with the clinical presentation to validate the findings and proceed with management accordingly.

The simultaneous occurrence of pneumoperitoneum, pneumoretroperitoneum, pneumomediastinum, and pancreatitis following ERCP is exceedingly rare. Prior cases have outlined similar occurrences, with variations in clinical presentation and management compared to ours [9–11].

Fujii et al.'s [9] study focused on managing complications related to anastomotic stricture post-ERCP, iatrogenic subcutaneous emphysema, and pneumothorax in a 73-year-old female with a history of gastrointestinal surgeries. Jha et al. [10] reported cases involving complications such as pneumothorax and subcutaneous emphysema during the ERCP procedure in a 65-year-old hypertensive female and a 25-year-old woman. In a case reported by Ferrara et al. [11], an elderly male patient experienced postprocedural complications, including hypotension, tachycardia, decreased oxygen saturation, and the development of various complications following ERCP. Notably, the absence of underlying significant comorbidities and successful conservative management distinguished our case from previously reported cases.

Two main classification systems exist to categorize ERCP-related perforations: the Howard classification system and the Stapfer classification system. The Howard classification system groups perforations into three categories based on the mechanism of ERCP-related perforation [7]. Group I is associated with guidewire perforations, group II with periampullary

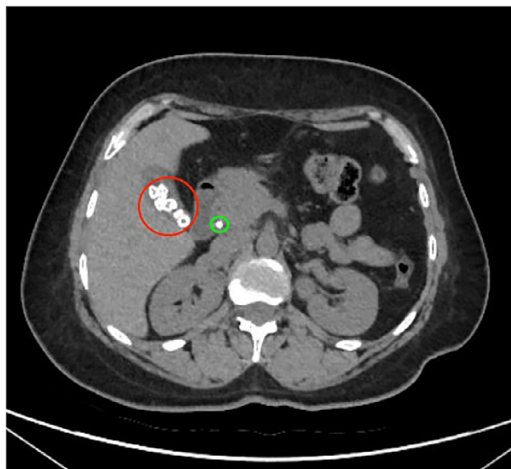


Fig. 3 – Follow-up axial CT scan showing the resolution of pneumoperitoneum and pneumoretroperitoneum, while cholelithiasis remains unchanged (red circle), and the in-place biliary stent is still observed.

perforations, and group III with duodenal perforations. On the other hand, the Stapfer classification system identifies 4 types of perforations based on the severity and anatomical location of the injury [3]. Type I perforations are caused by the endoscope itself, tend to be significant and are usually intraperitoneal. Type II is caused by manipulation of the ampulla during sphincterotomy or other therapeutic measures, and it is the most common type of injury [12]. Type III corresponds to bile or pancreatic duct injury, comparable to Howard group I, since most of these perforations are caused by guidewire instrumentation. Type IV is identified by the presence of retroperitoneal air alone.

It is important to note that retroperitoneal air can be observed on CT scans in up to 29% of asymptomatic patients after an ERCP, irrespective of the procedure's duration [13,14]. This phenomenon is caused by using compressed air during the procedure to maintain the patency of the duodenum. Therefore, clinicians and healthcare professionals should be aware of the different classification systems for ERCP-related perforations to ensure prompt and effective management of any potential complications.

In analyzing our case, the presentation aligns with a Stapfer type III perforation characterized by bile or pancreatic duct injury during the ERCP procedure. This classification resonates with the development of acute pancreatitis, confirmed by elevated serum amylase and lipase levels, as well as the radiological evidence of pneumoperitoneum, pneumoretroperitoneum, and pneumomediastinum on the CT scan. Identifying the distal CBD or duodenal perforation as the probable source of the observed pneumoretroperitoneum and pneumoperitoneum is consistent with the mechanism of a type III perforation [3].

The management of duodenal perforation post-ERCP is a topic of debate. Howard et al. [7] reported that conservative management was successful in 36 out of 40 patients. The group suggests a risk stratification approach based on the mechanism of injury, location of perforation, and time of di-

agnosis to determine if operative intervention is necessary. Stapfer et al. [3] reviewed 14 cases of ERCP-related perforations and concluded that, for stable patients, conservative management was more effective than surgical intervention. Operative management is individualized but typically involves draining leaked contents, repairing duodenal defects, and performing a cholecystectomy [3].

Understanding the nuances of the Stapfer classification, particularly type III perforations, is crucial in guiding the management approach. The conservative management employed in our case, including NPO, nasogastric tube insertion, intravenous fluids, pain relief medication, and broad-spectrum antibiotics, was in line with the established recommendations for stable patients with ERCP-related perforations.

Comparing our case with previously reported instances, it becomes evident that our patient's clinical profile and successful conservative management without surgical intervention distinguish it as a unique representation of a type III perforation following ERCP. The successful conservative management without surgical intervention further underscores the significance of timely diagnosis and appropriate intervention in ensuring favorable patient outcomes.

We thank the patient and medical team for contributing to this ERCP complication report. Our aim is to improve awareness and management strategies for such events, ultimately enhancing patient safety and outcomes.

Conclusion

This unique case underscores the intricate nature of complications arising from ERCP, highlighting the simultaneous occurrence of acute pancreatitis, pneumoperitoneum, pneumoretroperitoneum, and pneumomediastinum. The successful conservative management without resorting to surgery emphasizes the pivotal role of prompt diagnosis and tailored intervention in optimizing patient outcomes. As we navigate the complexities of ERCP-related perforations, this case serves as a poignant reminder of the need for heightened awareness, meticulous classification, and individualized patient care. By sharing such experiences, we contribute to the collective knowledge, fostering a deeper understanding of these rare events and ultimately advancing the safety and well-being of patients undergoing ERCP procedures.

Author contributions

All authors had access to the data and a role in writing this manuscript.

Data availability statement

Not applicable.

Patient consent

Written informed consent was obtained from the patient for their anonymized information to be published in this article (case report).

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