Radiology Case Reports 17 (2022) 987–990

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Case report

Pancreaticopleural fistula in children: Report of 2 cases☆

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ARTICLE INFO

Article history:
Received 15 December 2021
Revised 1 January 2022
Accepted 5 January 2022

Keywords:
Pancreaticopleural fistula
Fistulo-jejunostomy
Cystojejunostomy

ABSTRACT

Pancreaticopleural fistula is an extremely rare complication of pancreatic duct injury. The reported treatments include conservative approaches, such as pleural drainage, and interventional approaches, such as sphincter stenting via endoscopic retrograde cholangiopancreatography and surgery. However, no specific consensus treatment has been defined. We present 2 cases of pediatric patients with pancreaticopleural fistulas due to pancreatic trauma and pancreatitis that were successfully treated surgically. The most prominent symptom in both cases was dyspnea caused by pleural effusion. Thoracoabdominal computed tomography scans showed large pleural effusions and visible fistulas from the pancreatic duct to the thoracic cavity through the esophageal hiatus and aortic hiatus. Following unsuccessful conservative treatment using pleural drainage, the 2 patients underwent surgical fistulo-jejunostomy and cystojejunostomy. Both patients were stable and were discharged on postoperative days 10 and 12. Conservative treatment for pancreaticopleural fistula often fails, and a surgical approach, such as fistulo-jejunostomy and cystojejunostomy, can serve as an efficacious management strategy when conservative treatment fails.

Introduction

Pancreaticopleural fistula is a rare complication that can present in both adults and children. In 2019, Yu et al. identified only 9 cases across 7 reports in the English-language literature during the past 30 years [1]. In 2020, Zhang et al. identified only 22 cases across 6 reports in the Chinese-language literature and 8 reports in the English-language literature since 1980 [2]. Pancreaticopleural fistulas are

☆ Competing Interests: The authors have declared that no competing interests exist.

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https://doi.org/10.1016/j.radcr.2022.01.007

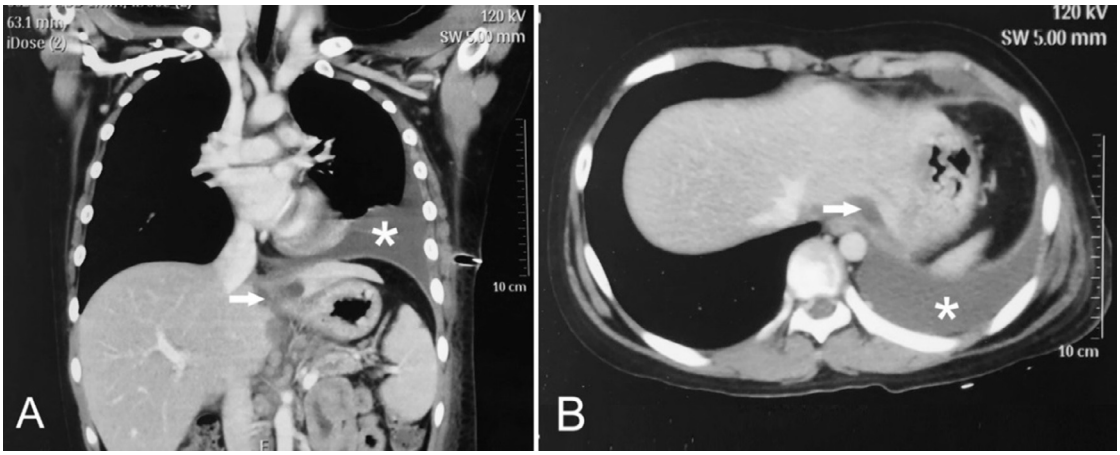

Fig 1 – Right pleural effusion (asterisks) and pancreaticopleural fistula (arrows) on the coronal (A) and axial planes (B) of thoracoabdominal computed tomography scans performed for Patient 1.

diagnosed when the pleural fluid contains high levels of pancreatic enzymes or imaging reveals the presence of a fistula between the pancreatic duct and the pleura, causing pleural effusion. The reported number of cases presenting with this complication is minimal, and no consensus regarding the best course of management for this condition exists. Reported treatments include conservative treatments, such as pleural drainage, and interventional treatments, such as endoscopic retrograde cholangiopancreatography (ERCP) sphincter stenting or surgery [3,4]. In this report, we present 2 cases of this rare entity and discuss aspects of diagnosis and treatment.

Case description

Case 1

A 13-year-old boy experienced a pancreatic duct disruption (grade 3 pancreatic trauma) due to a motorbike accident 4 months prior to admission to our hospital. At the time of the trauma, the patient was treated with conservative treatment at a provincial hospital and was stably discharged after 1 month. Three months after discharge, the patient was re-admitted to the same provincial hospital with abdominal pain and chest tightness. The patient was transferred to our center with a diagnosis of left pleural effusion and pancreatic pseudocyst. At the time of admission, the patient had stable vital signs and good nutritional status (148 cm, 43 kg). A thoracoabdominal computed tomography (CT) scan showed a large left pleural effusion causing atelectasis, mild dilatation of the caudal pancreatic duct, and a pancreatic fistula. The fistula had a diameter of 8-12 mm, originated from a duct at the pancreatic body, passed through the aortic hiatus anterior to the aorta, and opened into the left pleural cavity (Fig. 1). Initial treatment consisted of left pleural tube drainage to relieve dyspnea and antibiotics (third-generation cephalosporin and metronidazole). Pleural fluid tests showed elevated amylase and lipase concentrations of 27,427 and >60,000 IU/L, respectively. Blood tests showed normal C-reactive protein (CRP; 3.2 mg/L) and increased serum amylase/lipase (647/375 IU/L) levels. Conservative treatment continued for 14 days, at which time point, 100-150 mL/d of turbid yellowish pleural fluid indicated the presence of a secondary infection in the pleural cavity. The patient's blood culture came back positive for *Burkholderia cepacia*. Antibiotic treatment was escalated to pythinam and vancomycin, and surgery was indicated. The patient lost 5 kg during 20 days of hospitalization.

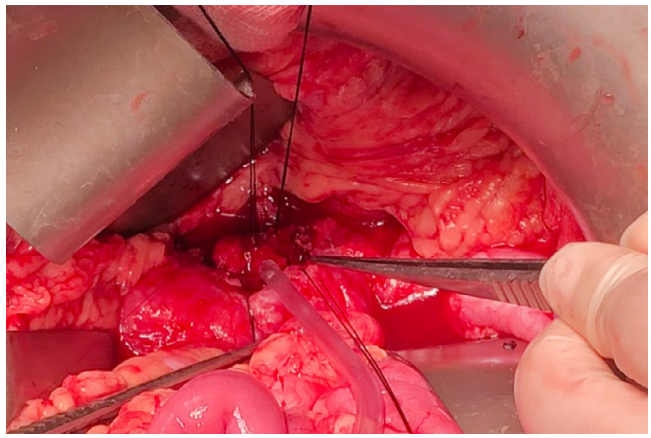

Fig. 2 – Intraoperative image of the pancreaticopleural fistula in Patient 1.

On day 20 after hospital admission, the patient underwent video-assisted thoracoscopic surgery-decortication with simultaneous pancreatic fistulo-jejunostomy using a Roux-en-Y jejunal limb. The pancreatic fistula was debrided to the body of the pancreas, where it communicated with the main pancreatic duct, followed by the performance of a fistulo-jejunostomy (Fig. 2). The postoperative course was uneventful, and the patient was discharged from the hospital with normal serum pancreatic enzymes and resolved pleural effusion on postoperative day 12. No further symptoms were reported at the 6-month follow-up.

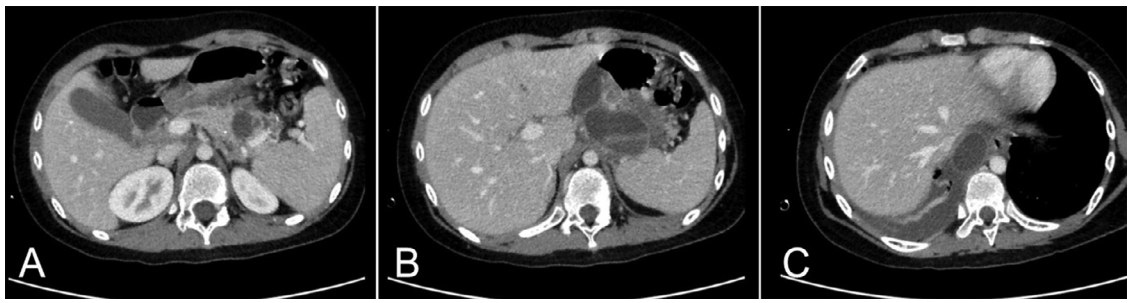

Fig. 3 – Thoracoabdominal computer tomography scan of Patient 2 showed a pseudocyst originating from the tail of the atrophic and calcified pancreas (A). The abdominal proportion (B) and the mediastinal proportion of the pseudocyst (C).

The fistula at the pancreatic body (forceps head) was tractioned after debridement and irrigation.

Case 2

A 13-year-old boy presented with a history of acute dyspnea and chest tightness that became intolerable over a 3-day period. The patient was transferred to us from a provincial hospital in stable condition with good nutritional status (150 cm, 42 kg). Chest x-ray showed a large right pleural effusion. Tube placement was used to drain 1000 mL of brownish-red fluid, which promoted immediate relief. Fluid amylase and lipase levels were 1260 and 4696 IU/L. Cell block analysis revealed no abnormal cells. High levels of pancreatic enzymes in the pleural fluid suggested effusion related to the pancreas, although abdominal signs and symptoms were unremarkable. CT scan was indicated and depicted a 35 × 75 cm pancreatic pseudocyst originating from the tail of the atrophic and calcified pancreas. A fistula, trajecting along the esophagus, connected 2 proportions of the pseudocyst: the mediastinal part and the abdominal part (Fig. 3). The diagnosis of pancreaticopleural fistula, pancreatic pseudocyst, and chronic pancreatitis was confirmed. Blood tests showed an increase in CRP (54.4 mg/L), amylase (296 IU/L), and lipase (510 IU/L). The patient was treated conservatively with pleural drainage, antibiotics, and pancreatic enzymes and was stabilized after 7 days, at which time the tube was removed due to the small flow volume of 20 mL/per day. Five days after removal, the pleural fluid reaccumulated, causing dyspnea, fever, and chest pain. The refractory effusion and infection resulted in an indication for surgery. During the conservative treatment period, levofloxacin was administered due to a *Burkholderia cepacian*–positive blood culture. The patient lost 3 kg during the 25 days of admission.

Intraoperatively, the pancreatic pseudocyst and the fistula were irrigated. The cyst was drained to a Roux-en-Y jejunal limb. The patient was discharged on postoperative day 10, with no further symptoms reported at the 3-month follow-up.

Discussion

The etiology of pancreaticopleural fistula in children can be pancreatitis, pancreatic trauma, or malformation of the pancreatic duct. In the 22 retrospective cases reviewed by Zhang et al., the most common cause (77.3%) was chronic pancreatitis, followed by pancreatic injury (18.3%) [2]. Our cases in this report were diagnosed with chronic pancreatitis and pancreatic trauma, represent the 2 most common causes of pancreaticopleural fistula. A traumatic antecedent in Patient 1 suggested a diagnosis of pancreatic involvement at the time of admission. However, Patient 2 presented with chronic pancreatitis with no suggestion of abdominal symptoms. The omnipresent respiratory signs could have resulted in a misdiagnosis without the laboratory results reporting amylase and lipase levels. Yu et al. discussed a similar finding in their report [1].

The diagnosis of pancreatic fistula is suspected when pancreatic enzymes are elevated in the pleural fluid [1–4]. Yang et al. defined the concentration threshold as amylase ≥1000 IU/L [3] and suggested that only pancreatic fistulas could increase amylase levels detected in the pleural fluid above 50,000 IU/L. However, no consensus for a specific threshold has been established, and such a threshold is rarely discussed in the literature [1,3]. In our opinion, the elevation of enzymes on fluid analysis represents a suggestive factor, and the identification of an anatomical connection through imaging remains necessary to confirm the diagnosis.

The performance of a CT scan or magnetic resonance imaging (MRI) is necessary to identify the entity underlying this disease [2,5,6]. In most cases, these imaging modalities will allow for fistula detection, in addition to associated lesions, such as pancreatic pseudocyst, pancreatic ductal malformation, pancreatic duct rupture, and chronic pancreatitis with atrophy and calcifications [3]. The fistulas from the pancreatic duct or pancreatic pseudocyst leak into the pleura through a natural hiatus between the thoracic and abdominal cavities. Either the left or right thorax can be affected, depending on the trajectory of the fistulas [2,7,8].

Currently, no established consensus or guideline exists regarding the treatment of pancreaticopleural fistulas in children, and evidence-based treatment recommendations are limited to case reports, which primarily involve adults [3]. Most authors choose conservative medical treatment for 2-4 weeks, followed by stenting ERCP or surgery after failure. Yu et al. reported a failure rate of 57.1% (4/7 cases) for conservative treatment. Many others have reported cases in which conservative treatment failed and required ERCP or surgery [3,5,7]. Both of our cases were initially and unsuccessfully treated

with conservative approaches, which ultimately failed due to persistent effusion and infection (Patient 1) or the rapid re-accumulation of fluid causing dyspnea (Patient 2). Due to the unavailability of stenting ERCP at our center, surgery was indicated in both cases. Although ERCP is reported to be a useful diagnostic and treatment tool, the success reported rate for stenting ECRP has varied, ranging from 50% reported by Wronski et al. [9] to 80% in other studies [1,2].

The key to a successful surgical approach is to treat the pancreas and re-establish the pancreatic juice flow. In addition to fistulo-jejunostomy and cystojejunostomy, authors have described the use of other procedures, such as pancreaticojejunostomy (Puestow procedure) or the removal of the pancreas tail [3,9–11]. Surgery should be performed promptly, as delayed response to the failure of conservative treatment can lead to malnutrition, catheter infection, sepsis, and other complications [11]. Our strategy was to perform surgery as soon as the infection was adequately controlled.

Conclusion

Pancreaticopleural fistula is a rare complication of pancreatic ductal injury with various etiologies. Although the disease originates from the pancreas, physicians should be aware that the predominant presentation includes respiratory symptoms due to pleural effusion. The biochemical testing of pleural fluid to detect the concentrations of amylase and lipase should be routinely performed in patients with effusion, regardless of abdominal signs. Elevated pancreatic enzymes in the pleural fluid and CT scans or MRI images can confirm the diagnosis and identify associated pancreatic lesions. Conservative treatment often fails, and surgical fistulo-jejunostomy or cystojejunostomy is an effective method for the treatment of pancreaticopleural fistula that fails to respond to conservative treatment.

Patient consent

Informed consent for patient information to be published in this article was obtained.

Authors' contributions

Tran TT and Pham NT contributed equally to this article as first authorship. All authors read and approved the final manuscript.

Data availability statement

All data generated or analyzed during this study are included in this article. Further inquiries may be directed to the corresponding author

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