CASE REPORT

An unexpected cause of pleural effusion in paediatric emergency medicine

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SUMMARY

Pancreaticopleural fistula (PPF) is an uncommon complication of chronic pancreatitis leading to a large and recurrent pleural effusion. Since the patients presented predominantly with respiratory symptoms, diagnosis and treatment were often delayed. We describe a child who was admitted to our paediatric emergency department with an acute onset of dyspnoea and unilateral massive pleural effusion caused by PPF. Multidetector CT is an easily accessible method that is able to show both the thoracic and abdominal findings non-invasively. The clinical and imaging features of this unusual entity are discussed.

BACKGROUND

The diagnosis of pancreaticopleural fistula (PPF) is often delayed because respiratory rather than abdominal symptoms predominate.^{1–5} In a majority of cases, the presenting findings are large, recurrent pleural effusion, dyspnoea and cough. Multidetector CT (MDCT) is an easily accessible method that is able to show both the thoracic and abdominal findings non-invasively.^{1–4}

CASE PRESENTATION

A 5-year-old girl was admitted to our paediatric emergency department with an acute onset of dyspnoea. She had a history of blunt abdominal trauma 2 years previously. She was complaining of intermittent abdominal pain for the last 6 months. On physical examination, she was found to be dyspnoeic and tachypnoeic and having dullness to percussion with decreased breath sounds on the right hemithorax. Other aspects of the physical examination were unremarkable.

INVESTIGATIONS

The routine haematology and biochemistry parameters were within normal limits except for the mildly elevated white blood cell count ($12.9 \times 10^9/l$) and erythrocyte sedimentation rate (42 mm/h). The chest x-ray revealed a large right-sided pleural effusion (figure 1A). Diagnostic thoracentesis exhibited exudates with a pleural fluid amylase 1200 U/l (serum amylase, 334 U/l). Cultures of pleural fluid were negative for aerobic, anaerobic and fungal agents. Also, the PCR tests were negative for tuberculosis and no malignant cells were observed at the cytological examination.

Abdominal ultrasonography (US) was performed in order to investigate the cause of intermittent abdominal pain. US depicted a mildly dilated pancreatic duct and a pseudocyst formation adjacent to the

Figure 1 (A) Chest radiograph on admission showing a large right-sided pleural effusion. (B) Diminished pleural effusion during treatment. (C) Significantly increased aeration of the right lung after chest tube removal.

pancreatic head. Abdominal MDCT identified a dilated irregular pancreatic duct, parenchymal atrophy with several pseudocysts ranging in size from 8 to 20 mm and also a fistulous tract originating from a pseudocyst and extending to the right hemithorax (figure 2). According to the radiological appearance, pleural effusion was thought to be secondary to PPF as a complication of chronic pancreatitis.

TREATMENT

A chest tube was placed in the right hemithorax. In addition, total parenteral nutrition, intravenous infusion of octreotide and appropriate antibiotic treatment were started. A follow-up CT scan revealed a decrease in the size of the fistula and pseudocysts and also in the amount of the pleural fluid at the end of the second week of treatment. The chest tube was removed when the pleural effusion resolved (figure 1B,C).

Figure 2 Coronal-oblique (A) and sagittal-oblique (B) reformatted CT scans showing the fistulous tract arising at the level of the head of the pancreas and extending through the right pleural cavity.

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OUTCOME AND FOLLOW-UP

Medical conservative treatment resulted in spontaneous closure of the fistula and surgical intervention was not required. She was discharged from the hospital on the 50th day of hospitalisation.

DISCUSSION

Pleuropulmonary complications such as atelectasis, pneumonitis, adult respiratory distress syndrome and pleural effusion are relatively frequent in patients with acute pancreatitis.^{1 4 5} Small, left-sided and self-limited pleural effusions are probably the result of a sympathetic, chemically induced diaphragm-pleural inflammatory process and occur in 4–17% of patients with acute pancreatitis.^{1 4} They resolve spontaneously as the pancreatic inflammatory process subsides.⁴ However, the effusion in chronic pancreatitis and PPF is often large and recurrent. It is usually left sided, but may even be right sided or bilateral.⁵

Secondary to the chronic inflammation of the pancreas, the main pancreatic duct is obstructed and ruptured in the subacute state.^{6 7} Pancreatic duct disruption develops secondary to alcoholic pancreatitis or trauma in adults and the formation of pseudocysts during the course of chronic pancreatitis in children.² If the ductal leakage is anterior, pancreatic secretions flow freely into the abdominal cavity so pancreatic ascites occurs.^{6 7} If it is posterior, the retroperitoneal pancreatic fluid tends to go towards the mediastinum because of the transdiaphragmatic pressure gradient between the abdominal and pleural cavities. The fluid uses the oesophageal or aortic hiatus as a migration path.^{2 6 7} More rarely, it passes via direct penetration through the dome of the diaphragm ignoring the normal fascial planes.^{2 4} A mediastinal pseudocyst occurs when the secretions are contained by the pleura. Rupture of the pleura results in a chronic large pleural effusion, in contrast to the self-limited, small pleural effusion of pancreatitis.⁶

The pleura fluid is exudative and may contain blood cells, as well as a high concentration of protein. A high amylase level in the pleural fluid is a typical finding of pleural effusions associated with chronic pancreatitis, but this does not always confirm a PPF.^{1 2 5} Acute pancreatitis, oesophageal perforation, parapneumonic effusion, and lung and pancreatic malignancy are other causes of this situation. As it is different from others, the amylase levels of pleural fluid reaches high values, such as 50 000 IU/l in PPF. Pleural fluid contains a salivary-type amylase isoenzyme in non-pancreatic pathologies such as oesophageal perforation and lung cancer.³ However, pancreatic pleural effusion is characterised by high amylase content, predominantly pancreatic-type amylase isoenzyme.^{2 3}

Owing to the posterior rupture of the pancreatic duct and decompression of enzymes into the mediastinum, patients have few abdominal signs and symptoms. The presenting complaints are generally by thoracic origin such as dyspnoea, chest pain and cough. Patients with pancreaticobronchial fistulas may also have recurrent lobar lung consolidations.³

Imaging findings can be helpful in diagnosis if the amylase level is moderately elevated. Although transabdominal ultrasound has been reported to be useful for diagnosing PPF,⁸ an overlying bowel gas artefact and inadequate respiratory cooperation are the limitations of US examination especially in the paediatric age group. Signs of chronic pancreatitis or the presence of pseudocysts can be seen with US. But most of the time it is not possible to visualise a fistulous tract.

MDCT is very useful for imaging findings of chronic pancreatitis and it is also excellent in showing pancreatic parenchymal changes, pseudocysts, calcifications, peripancreatic fat infiltration

and the extent of fluid accumulations.^{1 4} It can clarify the site of fistulisation and give details of anatomical relations of the fistula with other organs. The fistulous tract extending from the abdomen to the pleura, bronchi or mediastinum can be revealed. The extension of pseudocysts to the pleura and mediastinum and inflammation extending from the pancreas to the crus of the diaphragm are suggestive findings of pancreaticothoracic fistula.³ The ability of MDCT to detect the fistula tractus has been variable, ranging from 0% to 100% in previous case series depending on the amount of fluid present in the fistula at the time of examination.³ CT has the advantage of being non-invasive and the capability of providing three-dimensional images.

Endoscopic retrograde cholangiopancreatography (ERCP) has been used for the diagnosis of PPF. The accuracy of ERCP in diagnosing a pancreaticothoracic fistula is variable.³ It can demonstrate a pancreatic ductal anatomy and fistulous tract directly but sometimes fails to show the thoracic portion of the fistula.^{2 3} ERCP is an invasive procedure and also has a small but substantial complication rate. Acute pancreatitis, sepsis and bleeding are among these complications. ERCP is not always available and also difficult to perform in paediatric cases.

Tube thoracostomy and suppression of pancreatic enzymes should be applied as part of the initial conservative management.² Invasive procedures such as pancreatic duct stenting or operative treatment are necessary where conservative approach fails.⁴ Our patient was treated conservatively with octreotide, total parenteral nutrition and appropriate antibiotherapy and she responded well.

In conclusion, although it is extremely less common, chronic pancreatitis and PPF should be considered in differential diagnosis of massive pleural effusion in patients who come to the emergency department with acute respiratory distress. Since the patients presented predominantly with respiratory symptoms, diagnosis and treatment were often delayed. The rich amylase content (predominantly pancreatic-type amylase isoenzyme) of the pleural fluid is an important diagnostic tool. MDCT of the chest and abdomen is valuable in demonstrating chronic pancreatitis and the presence of fistulisation with the advantage of being non-invasive and the capability of providing three-dimensional images.

Learning points

- Pancreaticopleural fistula (PPF) is an uncommon complication of chronic pancreatitis leading to large and recurrent pleural effusion.
- Diagnosis of PPF is often delayed because respiratory symptoms rather than abdominal predominate.
- Multidetector CT is an easily accessible method that is able to show both the thoracic and abdominal findings, as well as the fistulous tract non-invasively.

Competing interests None.

Patient consent Obtained.

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