

Case report

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ACUTE ASCENDING MEDIASTINITIS AND NECK PHLEGMON CAUSED BY PANCREATIC PSEUDOCYST: CASE REPORT

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Acute mediastinitis is one of the most severely known infections with high mortality rate. Mediastinitis occurs most commonly as a descending infection, spreading from oral cavity or pharynx. The most common thoracic complications of pancreatic disease are reactive pleural effusion and pneumonia, while rare complications include pancreaticopleural fistula with massive pleural effusion or hemothorax and extension of pseudocyst into the mediastinum. Only 50 cases of mediastinal extension of pancreatic pseudocyst have been published in the last 75 years with only one case of neck involvement reported. We report a case of acute respiratory distress syndrome followed by toxic enzymatic hydropneumothorax, mediastinitis and neck phlegmon caused by pancreatic pseudocyst fistula in 45 years old man. After initial diagnostics emergency operation was performed by multidisciplinary team. Ascending mediastinitis caused by pancreatic pseudocyst required a prompt surgical intervention. Early detection and an aggressive approach are crucial. Treatment should be a combination of surgical debridement and broad-spectrum antibiotics followed by adequate supportive care.

Key words: *mediastinitis, pancreatic pseudocyst, neck phlegmon*

Prikaz slučaja

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AKUTNI ASCEDENTNI MEDIJASTINITIS I FLEGMONA VRATA UZROKOVANI PSEUDOCISTOM PANKREASA: PRIKAZ SLUČAJA

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Akutni mediastinitis spada u grupu najtežih infekcija sa visokom stopom mortaliteta. Najčešće nastaje kao posledica descendentnog širenja infekcije iz usne duplje ili farinksa. Najčešće torakalne komplikacije bolesti pankreasa su reaktivni izlivi ili pneumonije, dok se komplikacije poput pankreatično-pleurane fistule praćene masivnim izlivom ili hemotoraksom i ekstenzijom pseudociste u mediastinum retko sreću. Samo 50 slučajeva mediastinalnog širenja pseudociste je objavljeno u poslednjih 75 godina, a kod samo jednog sa širenjem u predeo vrata. Prikazan je slučaj akutnog respiratornog distresa sa razvojem enzimskog hidropneumotoraksa, mediastinitisom i flegmonom vrata uzrokovan pseudocistom pankreasa kod bolesnika starog 45 godina. Nakon inicijalne dijagnostike bolesnik je podvrgnut urgentnoj operaciji koju je izveo multidisciplinarni tim. Ascendentni mediastinitis uzrokovan pseudocistom pankreasa zahteva hitno hirurško lečenje. Rana detekcija i agresivni pristup u lečenju su ključni. Terapija podrazumeva kombinaciju hirurškog debridmana uz antibiotike širokog spectra i adekvatnu suportivnu terapiju.

Ključne reči: medijastinitis, pseudocitsa pankreasa, flegmona vrata

Introduction

Acute mediastinitis is one of the most severely known infections with high mortality rate ranging between 14-42% (1). Most commonly, mediastinitis occurs as a descending process arising from oral cavity or pharyngeal infection spreading across vascular sheaths or prevertebral fascia to visceral or anterior mediastinum. The most common thoracic complications of pancreatic disease are reactive pleural effusion and pneumonia, while rare complications include thoracic conditions, such as pancreaticopleural fistula with massive pleural effusion or hemothorax and extension of pseudocyst into the mediastinum (2). Only 50 cases of mediastinal extension of pancreatic pseudocyst have been published since 1951. Mediastinitis as a complication of pancreatic pseudocyst fistula is very rare and only one case of neck involvement has been reported (3).

Here, we report a case of acute respiratory distress syndrome followed by toxic enzymatic hydropneumothorax, mediastinitis and neck phlegmon caused by pancreatic pseudocyst fistula.

Case study

A 45-year-old man was admitted to emergency department in the state of severe septic shock, hypotensive (TA-95/58 mmHg, HR 105bpm) with redness and swelling of the neck, high fever (39.2°C), already on a mechanical ventilation. He had medical history of acute pancreatitis three years ago, followed by with bilateral pleural effusion. Otherwise, healthy man with no history of alcohol abuse or cholelithiasis. Deep neck infection was suspected. Lab analyses showed elevated blood amylase levels (4690 U/L, ref: 0 - 490) urine amylase level (17360 U/L, ref: 25-125), high leukocyte count ($28.1 \cdot 10^9/L$, ref: 4.0-9.0), increased level of C- reactive protein (261.4 mg/L, ref: 0.0-5.0), Cholesterol (4.2 mmol/L, ref: 3.90 - 5.20), Triglyceride (0,75 mmol/L, ref: 0,70 - 1.70), BMI (21,2 ref: 18,5-24.9).

CT scan of the neck, chest and abdomen was performed showing fluid accumulation in the posterior and visceral mediastinum, in both pleural spaces and in front of the prevertebral fascia of the neck (Figure 1, 2 and 3). Pancreas was inflamed with two pseudocyst located in the body, extending to posterior mediastinum. Size of pseudocysts was 76x47mm and 123x96mm respectively, both accumulating opaque dense fluid (Figure 4).

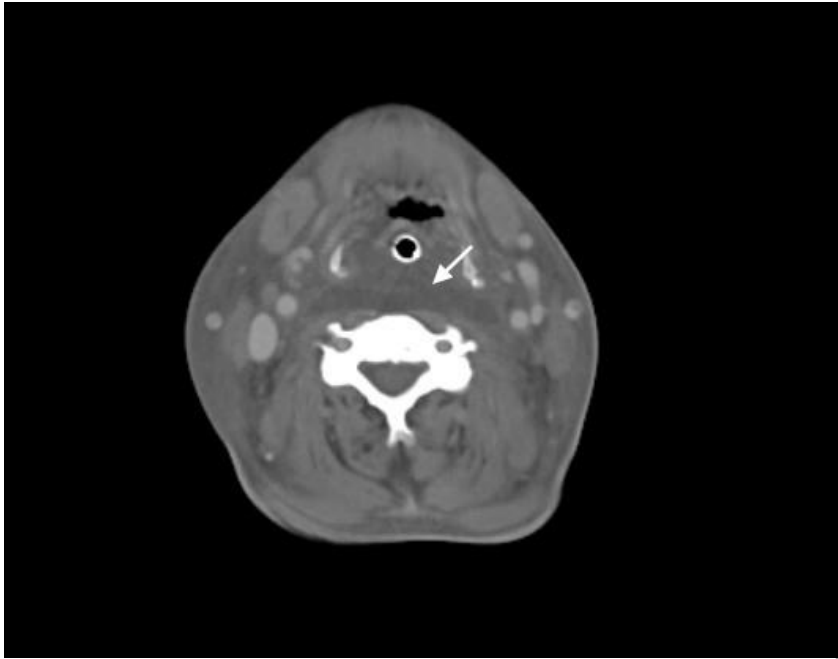


Figure 1. MSCT presentation of fluid collection in retropharyngeal space of the neck in front of prevertebral fascia (marked with an arrow)

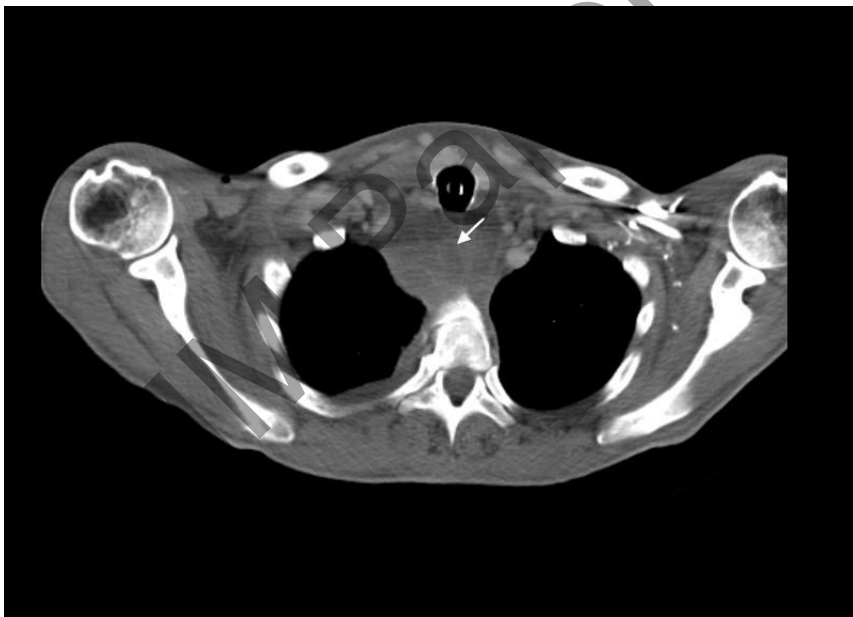


Figure 2. MSCT presentation of fluid collection in visceral mediastinum (marked with an arrow)

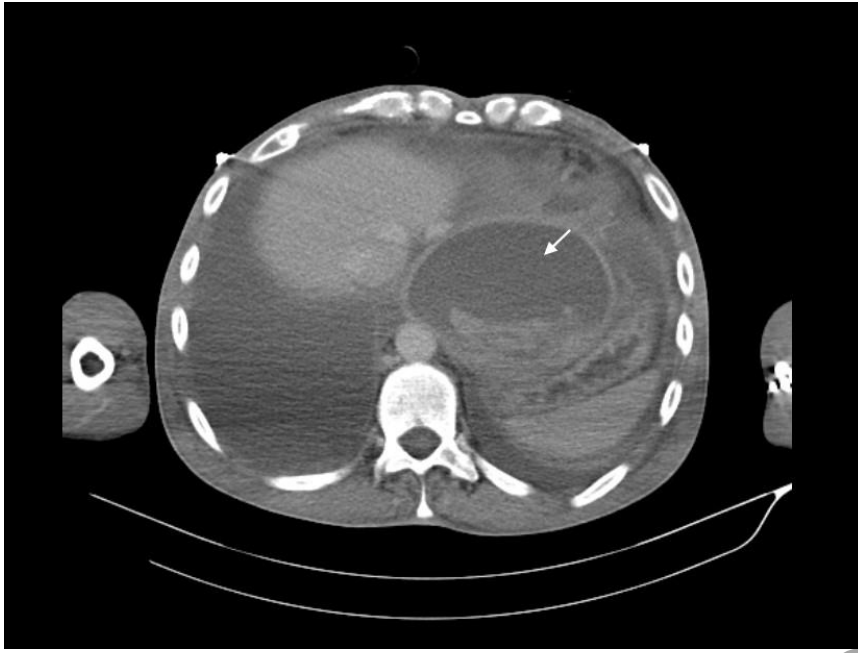


Figure 4. MSCT presentation of pancreatic pseudocyst (marked with an arrow)

Emergency operation was performed within one hour by a team of maxillofacial, thoracic and abdominal surgeon. The first step of the operation considered cervicectomy and drainage of the prevertebral space all the way to the thoracic inlet. Prevertebral space was full off thick, dark fluid which later showed elevated levels of pancreatic amylase (1339 U/L). Tracheostomy was performed. The next step was right anterior thoracotomy. Around 1000 ml of opaque fluid was evacuated from the pleural cavity and afterwards mediastinal pleura was widely opened. Debridement and drainage of mediastinum was performed with evacuation of the necrotic tissue and thick, dark fluid which later showed elevated levels of pancreatic amylase (18999 U/L). The operation was continued with upper median laparotomy which revealed pancreatic cysts extending to the posterior mediastinum. Simple surgical drainage of pancreatic cysts was performed followed by nutritive gastrostomy which was temporarily placed due to neck phlegmon.

The patient was transferred to the intensive care unit after the operation. The immediate postoperative recovery went slow with patient on mechanical ventilation. He was extubated on the 5th postoperative day with lungs showing full reexpansion and the absence of pleural complications (Figure 5). Parenteral nutrition combined with somatostatin treatment and gastrostomy rehydration was introduced. Neck, mediastinal, pleural and abdominal drains were removed in the following days. Lab results showed normalized levels of inflammation markers. Control CT scan showed no

sign of neck and mediastinal infection (Figure 6). Patient was transferred to standard care unit for further treatment with prolonged hospitalization due to impaired pancreatic function which was manifested by oscillations of blood glucose levels from severe hypoglycemia to moderate hyperglycemia.

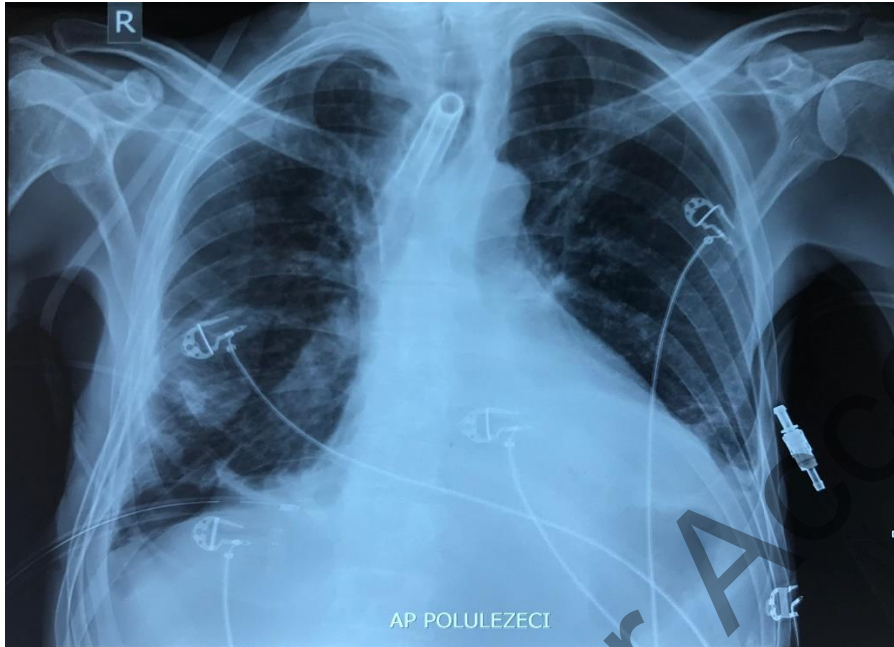


Figure 5. Postoperative X-ray showing reexpansion of both lungs and absence of pleural effusion



Figure 6. MSCT showing no sign of mediastinal infection

Unfortunately, four months after the operation patient had a new relapse of acute pancreatitis with severe multiple organ failure and absence of mediastinal complications. Lab analysis showed complete endocrine and exocrine pancreatic insufficiency. The outcome was fatal.

Discussion

Infections of the mediastinum are serious and associated with high morbidity and mortality. They may result from adjacent disease with direct extension, hematogenous spread, or direct introduction into the mediastinal space (4). Common causes of acute mediastinitis include surgical infection, esophageal rupture, and descending infection from the pharynx. Ascending infection from an intra-abdominal focus to the mediastinum is very rare. The clinical situation of ascending mediastinitis due to pancreatic pseudocyst is similar to the fulminant course of descending necrotizing mediastinitis (5). In this case chronic recurrent pancreatitis lead to formation of two pseudocysts with their extension to posterior mediastinum forming an active fistula. This subsequently lead to enzymatic mediastinitis with the enzymes extending to the retropharyngeal space. This is only the second case reported in literature with pancreatic fistula extending to deep fascial layers of the neck. Pancreaticopleural fistula should be suspected in patients with intense abdominal and chest pain followed by cough and hemoptysis. Opaque and dark fluid find in thoracocentesis should rise a suspicion of enzymatic pleuritic which can easily be confirmed on fluid analysis showing a high level of amylase. CT scan of chest and abdomen is mandatory and prompt surgical intervention is required.

Less invasive techniques, such as VATS, have become effective options for the treatment of mediastinitis by pleuromediastinal drainage with or without cervical drainage (6). Since our patient was in septic shock and due to massive neck phlegmon we opted for open surgery approach. Regarding pancreatic cysts CT, US or EUS guided percutaneous catheter drainage and endoscopic drainage, have gained popularity (7, 8, 9). When pancreatitis with pseudocyst is a result of pancreatic duct rupture, stent placement via transpapillary approach is considered (10).

Supportive treatment following surgery, with parenteral nutrition combined with or without somatostatin or its analogue, octreotide, has been shown to improve outcome in patients with mediastinal pancreatic fluid accumulation (11). We introduced somatostatin on first postoperative day which was continued until resolution of pancreatitis.

Our case showed an importance of multidisciplinary surgical approach. Cervicotomy and adequate necrectomy, debridement, lavage and drainage of the mediastinum, use of wide spectrum antibiotics and adequate supportive therapy in the setting of ICU, solved the severe and possibly fatal acute infection and patient did not die due to this complication of pancreatitis. Drainage of pseudocysts stopped the mediastinal infection recurrence in new onset of acute pancreatic infection.

However, pancreatitis is an inflammatory disease that results in a spectrum of mortality related to the severity of disease (12). Inflammatory cascade leads to the systemic manifestations of acute pancreatitis. It ultimately produces capillary permeability and endothelium damage with microvascular thrombosis that gives rise to multi-organ dysfunction syndrome (MODS) as the main cause of morbidity and mortality in the setting of acute pancreatitis (13).

Our patient was otherwise healthy man with no history of alcohol abuse or cholelithiasis. Some individuals have a genetic predisposition for pancreatitis and suffer from recurrent acute pancreatitis, progressing to chronic pancreatitis. Not surprisingly, the associated genes are involved in activating trypsinogen to trypsin. Many genes play a role in recurrent pancreatitis, e.g., cystic fibrosis transmembrane conductance regulator (*CFTR*) gene, cationic trypsinogen gene (*PRSS1*) gain of function mutations, mutations of trypsin degrading enzyme (CTCR) chymotrypsin C and (SPINK1) pancreatic secretory trypsin inhibitor. Furthermore, they are also involved in the increasingly acknowledged spectrum of diseases, from acute to chronic pancreatitis (14, 15, 16).

Conclusion

Ascending mediastinitis caused by pancreatic pseudocyst requires a prompt surgical intervention. Although very rare, infection can spread all the way to deep fascial layers of the neck. Early detection and an aggressive approach are crucial in reducing the high mortality rate infection. Treatment should be a combination of surgical debridement and broad-spectrum antibiotics followed by adequate supportive care. Acute pancreatitis is a severe disorder that still carries a high mortality rate due to multiorgan failure.

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