

## O P I S Y P R Z Y P A D K Ó W CASE REPORTS

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## A spontaneous resolution of mediastinal pancreatic pseudocyst compressing oesophagus and heart's posterior wall after total parenteral nutrition – a case report and literature review

## Samoistne wchłonięcie się torbieli rzekomej trzustki w śródpiersiu uciskającej na przełyk i tylną ścianę serca – opis przypadku i przegląd literatury

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lekasia87@wp.pl**S u m m a r y**

Pancreatic pseudocyst commonly complicates the course of acute and chronic pancreatitis. Very rarely pseudocysts can extend up to the mediastinum causing non-specific symptoms and posing diagnostic and therapeutic challenge. We report a case of a 34-year old male, with a history of chronic pancreatitis and alcohol abuse, who presented to the emergency department with severe abdominal pain, nausea, vomiting and dyspnea for the last 7 days. Laboratory results revealed leucocytosis, elevated level of CRP and pancreatic enzymes. He was admitted to the gastroenterology ward with a diagnosis of acute on chronic pancreatitis. Due to the suspicion of gastric outlet obstruction X-ray and CT-scan was performed. Large mediastinal pseudocyst compressing oesophagus and heart's posterior wall was found. EUS-guided drainage was not performed due to peptic ulcers in antrum, erosive gastritis, bulbar duodenitis seen at upper G.I. endoscopy and acquired coagulation disorder. Therefore we decided on implementing watchful waiting strategy which included total parenteral nutrition. After 26 days of conservative treatment resolution of symptoms was achieved and in a 3-month follow-up period with strict alcohol abstinence, spontaneous resolution of the pseudocyst was obtained. Our case emphasize that disappearance of mediastinal pseudocyst without invasive procedure is achievable and may be considered even in symptomatic patients. Written consent from the subject was obtained.

**S t r e s z c z e n i e**

Przebieg zarówno ostrego, jak i przewlekłego zapalenia trzustki może być powikłany wytworzeniem się torbieli rzekomej. Bardzo rzadko torbiele te mogą penetrować aż do śródpiersia, dając niespecyficzne objawy, a tym samym stanowiąc wyzwanie diagnostyczno-lecznicze. Prezentujemy przypadek 34-letniego mężczyzny z przewlekłym zapaleniem trzustki i zespołem zależności alkoholowej, który zgłosił się z powodu silnych dolegliwości bólowych brzucha z towarzyszącymi nudnościami, wymiotami oraz dusznością od 7 dni. W badaniach laboratoryjnych stwierdzono leukocytozę, podwyższony poziom CRP i enzymów trzustkowych. Chorego z rozpoznaniem zaostrzenia przewlekłego zapalenia trzustki przyjęto na Oddział Gastroenterologii. Z uwagi na podejrzenie niedrożności przewodu pokarmowego wykonano RTG przeglądowe jamy brzusznej, a następnie TK jamy brzusznej z kontrastem, które uwidocznili dużą torbiel rzekomą śródpiersia uciskającą na przełyk oraz tylną ścianę serca. Wykonano gastroskopię, uwiadczniając nadżerkowe zapalenie żołądka, owrzodzenia w antrum oraz zapalenie błony śluzowej opuszki dwunastnicy, co łącznie z istotnymi nabytymi zaburzeniami krzepnięcia skutkowało odstąpieniem od drenażu torbieli pod kontrolą EUS. Z tego względu zdecydowano o wdrożeniu strategii zachowawczej z włączeniem całkowitego żywienia pozajelitowego, co po 26 dniach leczenia spowodowało ustąpienie objawów, a w trzymiesięcznym okresie obserwacji w abstynencji doprowadziło do całkowitego wchłonięcia się torbieli rzekomej. Chcieliśmy wykazać, że postępowanie w torbieli rzekomej śródpiersia bez zastosowania procedur inwazyjnych jest możliwe i powinno się je rozważyć nawet u pacjentów objawowych. Uzyskano pisemną zgodę pacjenta.

## INTRODUCTION

Pancreatic pseudocyst is an encapsulated collection of fluid with a well defined inflammatory wall usually outside of the pancreas with no necrosis (1). It may develop as a common complication of acute pancreatitis (AP), chronic pancreatitis (CP) or pancreatic trauma. The incidence is reported to be about 30 to 40% (2) and alcohol-related etiology is implicated in up to 70% of cases (3). Peripancreatic region is a typical location for the pseudocysts. Nevertheless, rarely the disruption of pancreatic duct can lead to the leakage of the secretions through the paths of least resistance causing extensions to the spleen, liver, mediastinum, pelvis or kidneys (4, 5). Formation of mediastinal pseudocyst is rare and its exact incidence is being unknown (6). The etiology of mediastinal location is the most often related to ethanol-induced chronic pancreatitis (7). Due to uncommon location, often vast range of non-specific symptoms, pancreatic pseudocysts in atypical locations require high clinical vigilance which makes it a serious diagnostic and therapeutic challenge.

We report a case of a 34-year-old patient with a large mediastinal pseudocyst compressing oesophagus and heart's posterior wall which was formed as a result of acute on CP. No invasive procedures were performed and a spontaneous resolution of the pseudocyst observed after introduction of total parenteral nutrition. We present the successful watchful monitoring strategy in symptomatic patient and review available invasive procedures for mediastinal pseudocysts.

## CASE REPORT

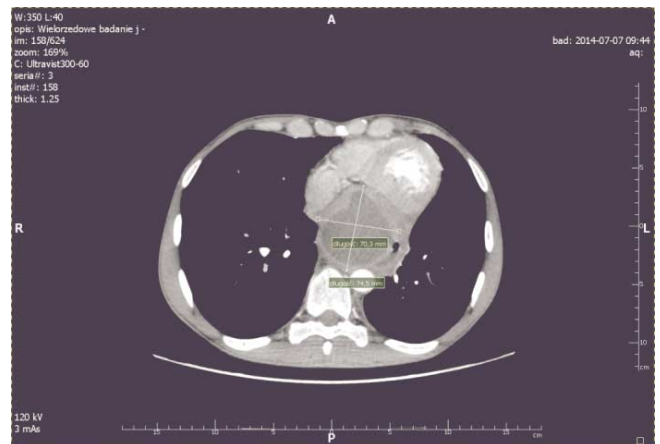
A 34-year-old Caucasian male, with a history of alcoholic CP, presented to the emergency department with a seven-day onset of severe upper abdominal and chest pain, dyspnea, nausea, vomiting and prominent dysphagia after solids and fluids. His past medical history revealed recurrent episodes of AP starting from 2008 with one of them complicated by a large pancreatic pseudocyst in peripancreatic region followed with successful percutaneous drainage. First exacerbation of CP was diagnosed in 2013, based on the widening of pancreatic duct and calcifications in the pancreatic body and tail revealed in CT scan. Eleven months prior to present episode he had developed chronic left side pleural effusion after acute pancreatitis. It resulted in undergoing multiple thoracocentesis procedures with pleural drainage and pleuroscopy with pleural and lung biopsy at that time.

On admission, laboratory results disclosed markedly elevated pancreatic enzymes: lipase – 1079 U/L (normal < 66 U/L), amylase – 332 U/L (normal: 22-80 U/L), urine amylase – 4047 IU/L (normal: 42-321 U/L) and CRP level – 47.16 mg/L, 191.25 mg/dL on the 8<sup>th</sup> day of hospitalisation (normal < 5 mg/L), leucocytosis – 13.84 G/L (normal 4-10 G/L), hypoalbuminemia – 29.52 g/L (normal 35-52 g/L), acquired coagulation disorder – PT 58% (normal 82-125%). Chest X-ray showed an oval mass in mediastinum with air-fluid level. On examination the

patient presented severe abdominal pain, cessation of peristalsis, hepatomegaly, tachycardia and suppressed vesicular sounds in the base of the left lung. Ultimately he was admitted to the gastroenterology ward with a diagnosis of acute on chronic pancreatitis. Contrast-enhanced computed tomography (CT) of chest and abdomen revealed large pseudocyst ranging from pancreatic head continuing up to mediastinum and left atrium with its estimated size around 78 x 70 x 145 mm compressing oesophagus and heart's posterior wall (fig. 1-3). In addition CT demonstrated left sided hydrothorax, possible inflammation around the aorta in the inferior mediastinum, inflammation in peripancreatic tissue, calcifications in pancreatic head, dilatation of pancreatic duct 8 mm in the body and up to 16 mm in the head (fig. 4).

In the course of treatment standard therapy for moderately severe phase of pancreatitis and total parenteral feeding for 15 days was implemented.

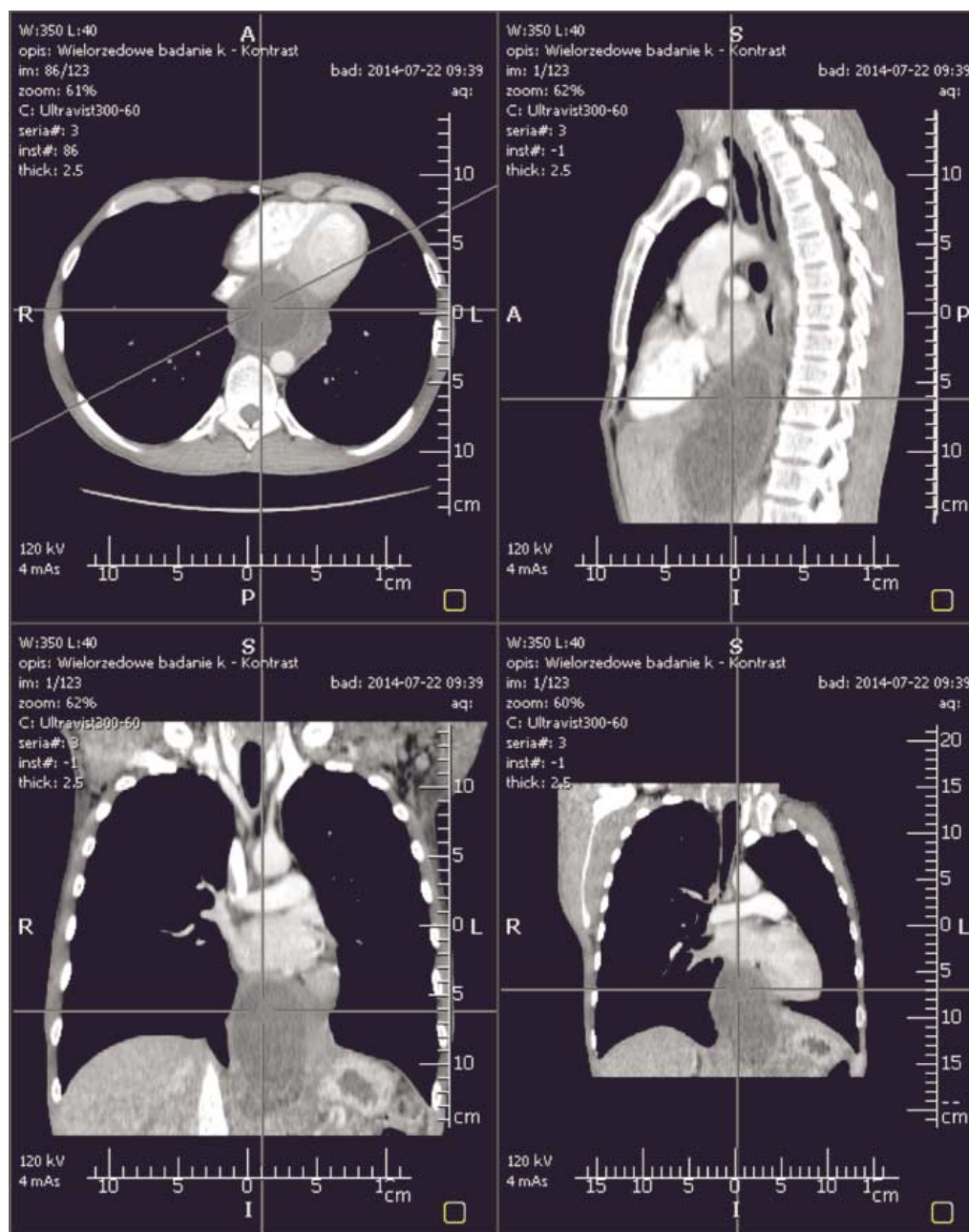
EUS-guided drainage and ERCP was planned but had to be postponed due to acquired coagulation disorder with a high risk of bleeding and the difficult transgastric approach due to the two peptic ulcers in antrum, erosive gastritis and bulbar duodenitis showed in upper G.I. endoscopy.



**Fig. 1.** Mediastinal pseudocyst compressing oesophagus and heart's posterior wall



**Fig. 2.** Pancreatic pseudocyst ascending through the diaphragm causing mediastinal extension



**Fig. 3.** Presentation of mediastinal pseudocyst in transverse, sagittal and coronal plane



**Fig. 4.** Massive calcifications and pancreatic duct dilatation in pancreatic head

By the time the coagulation results were satisfactory and optimal PPI treatment time for mucosal healing was reached, the patient felt improvement and bothersome symptoms were relieved. Therefore instead of invasive procedure the watchful monitoring strategy was introduced.

After 26 days of conservative therapy the patient was discharged from the hospital with complete relief of abdominal pain, nausea, vomiting, dyspnea and normalization of CRP and level of pancreatic enzymes. CT scan performed on discharge revealed a slight reduction of the size of mediastinal pseudocyst.

During a three-month follow-up period patient abstained from alcohol. Control CT-enhanced scan showed spontaneous resolution of mediastinal pseudocyst, post-inflammatory thickening of the periesophageal tissue, and partial regression of pancreatic infiltration.

Written consent from the subject was obtained.

## DISCUSSION

Mediastinal pseudocyst represents the rare complication of pancreatitis. Formation of mediastinal pseudocyst occurs when the pancreatic duct ruptures into the retroperitoneum and fluid enters into the thoracic cavity most commonly through the esophageal or aortic hiatus. Rarely, communication can occur via the foramen of Morgagni, the inferior vena cava hiatus or by direct erosion through the diaphragm (8). In our case the location of the fluid in the posterior mediastinum suggests extension most likely through esophageal or aortic hiatus.

Symptoms depend on the size, existence of pleural effusion often with high recurrence, compression of oesophagus or heart ventricles, which can result in chest pain, dysphagia, vomiting, dyspnea, arrhythmias or even with hemoptysis, acute respiratory distress (9), cardiac tamponade (10) and cardiogenic shock (11). Despite the location and size of the pseudocyst it can lead to serious life threatening complications which include: extension, internal hemorrhage, fistula, mediastinitis or rupture (12). The diagnosis requires not only high clinical vigilance due to presence of misleading symptoms, but also often careful radiological evaluation, beginning with chest radiogram, CT scan and continuing with endoscopic retrograde cholangiopancreatography (ERCP) or magnetic resonance cholangiopancreatography (MRCP) in order to assess ductal anatomy and communication.

Therapeutic strategies are as follows: watchful waiting monitoring, oral supplementation of somatostatin analogue and invasive procedures such as percutaneous drainage, endoscopic drainage and surgical procedure. Controversies exist not only regarding proper therapy but also intervention time.

The literature review shows only a few cases of spontaneous resolution of pancreatic pseudocyst in chronic pancreatitis requiring complete abstinence from alcohol (13, 14), bowel rest and total parenteral nutrition (15). Yasuda et al. reported complete disappearance of mediastinal pseudocyst after treatment with somatostatin analogue octreotide, which facilitated the closure of potential ductal fistula by inhibiting the production of pancreatic secretions (16).

Less invasive procedures such as nonsurgical drainage of concomitant pleural effusion resulting in resolution of mediastinal pseudocyst can still be effective in certain cases.

Drainage procedures are preferred options in the management of symptomatic mediastinal pseudocysts.

Nowadays endoscopic techniques are widely performed and are becoming the standard of care. ERCP with stent placement with or without transpapillary nasopancreatic drainage is used mainly when pancreatic duct communicates with pseudocyst disrupting in

the pancreatic head or body which can resolve both its mediastinal extension and pleural effusion (17-19). However the success rate of transpapillary technique in resolving pancreatic duct disruption was reported to be only 55% (20).

Newer techniques such as endoscopic ultrasound (EUS) are widely introduced. Impressive results of transgastric or transesophageal drainage of mediastinal pseudocyst are reported in the literature. Nevertheless this method is only available in large tertiary reference centers and requires significant experience of the operator. Possible complications are as follows hemorrhage, infection, esophageal stricture, post-procedure pancreatitis, stent migration and duodenal erosion.

Ultrasound-guided or more CT-guided percutaneous drainage are more accessible as a method. It can be both effective and combined with CT-guided stent placement (21). Complications of transcatheter external drainage include bleeding, infection, clogging of catheters with high-protein fluid, permanent fistula formation. Cyst recurrence and complication rate is higher than after EUS-guided drainage and is reported to be up to 20% (22).

Open surgical treatment is reserved for unstable patient with life-threatening presentations and complications such as rupture, infection, hemorrhage or failure of other techniques (23). Operating procedures that have been implemented vary from pancreatic resection to internal drainage like cystogastrostomy, cystojejunostomy or laparoendoscopic and thoracoscopic approach.

Fewer than 100 cases of the mediastinal pancreatic cyst expansion have been reported in the literature (24) that is why it is so hard to properly evaluate the safety and efficacy of techniques listed above.

Mediastinal pseudocysts although rare, could be a complication of common pancreatitis. The clue to success is remembering about the possibility of mediastinal location, especially in patient with alcohol-related chronic pancreatitis presenting with atypical symptoms. Due to potentially life-threatening complications and lack of specific management guidelines a multidisciplinary and individualized approach is so important. Choosing accurate treatment and appropriate time of intervention depend on clinical presentation, pseudocyst etiology, ductal anatomy, concomitant diseases and conditions and local experience. Nowadays endoscopic and surgical techniques used in pancreatic pseudocysts are quite satisfactory and still developing. Nevertheless overall percentage of recurrence and post-intervention complications are relatively high. In our case we implemented 15-days of total parenteral feeding and obtained pancreatic pseudocyst resolution. Therefore this report emphasize that mediastinal pseudocyst healing without invasive procedure is achievable and may be considered as a method of first choice even in symptomatic patients.

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