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Case Study

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A RARE CASE OF TUBERCULOUS EMPYEMA NECESSITANS COMPLICATING WITH PANCREATIC PSEUDOCYST AND CHRONIC PANCREATITIS RESULTING IN PANCREATICOPLEURAL FISTULA

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ABSTRACT

Tubercular empyema necessitans is a rare entity which occurs due to extravasation of pus loaded with tuberculous organisms from the chest wall with underlying heavily calcified pleura. Pancreaticopleural fistula is a complication of pancreatitis or pseudocyst occurring due to inflammation of pancreatic duct. We present a case of 26 year old male with pancreaticopleural fistula who presented with complaint of chest swelling which was diagnosed as tubercular empyema necessitans further it was complicated with chronic pancreatitis and pancreatic pseudocyst.

KEYWORDS: Empyema necessitans, pancreaticopleural fistula, chronic pancreatitis, pancreatic pseudocyst.

INTRODUCTION

Empyema necessitans is a rare long-term complication of poorly or uncontrolled empyema thoracis characterized by the dissection of pus through the soft tissues and skin of the chest wall.[1] The pus collection bursts and communicates with the exterior, forming a fistula between the pleural cavity and the skin.[1] Tuberculous empyema is a rare entity characterized by purulent pleural fluid that is loaded with tuberculous organisms on AFB stains.[2] Infected pleural effusion evolves into pus and a frank empyema. On rare occasions, a tuberculous empyema may produce an empyema necessitans where the empyema ruptures through the chest wall.[2] Indeed, TB is the most common cause of empyema necessitans.[2] This disease can be present in both immunocompetent individuals as well as immunocompromised patients. Due to the long latent period and enigmatic nature of the disease the diagnosis of this disease is difficult and disputed. Patients can present with enlarging mass on the chest wall which can be painful associated with chest pain, cough, dyspnea .Pus collection can also extend up to retroperitoneum, oesophagus and associated structures. Radiographically there can be presence of pleural effusion and radiographs shows pleural thickening. Computed tomography of chest shows loculated pleural collection or massive opacification of the affected side with calcified pleura.

A pancreatic pseudocyst as the name suggests is not a true cyst but it is a collection of debris and fluids which are rich in pancreatic enzymes either near or within the pancreas. The mechanism responsible for pleural effusion in patients with a chronic pseudocyst is the development of a direct sinus tract between the pancreas and the pleural space.[3,4] There is disruption of pancreatic duct which results in extravasation of the pancreatic fluid in the mediastinum and when it enters into pleural space, pancreaticopleural fistula results in massive pleural effusion which can be superadded with any infective pathogen.

Pancreaticopleural fistula (PPF) is a rare diagnosis that occurs approximately in 0.4% of patients with pancreatitis.[5] Most commonly left-sided effusions develop in patients with chronic pancreatitis who develop a pseudocyst with a sinus tract through the retroperitoneum into the pleural space. However mechanisms of formation of PPF either due to pancreatic pseudocyst or chronic pancreatitis is similar but due to absence of any significant abdominal symptoms results in delay in the diagnosis as patient complaint mainly of respiratory symptoms so initial efforts are directed mainly to rule out the thoracic pathology. A rare case of tubercular empyema necessitans complicating with pancreatic pseudocyst and chronic pancreatitis resulting in pancreaticopleural fistula is being reported.

CASE REPORT

A 26 years old male presented with complaints of breathlessness on exertion (MMRC grade II), cough with expectoration, fever, loss of appetite, loss of weight for 1 month duration with left side chest swelling on anterior chest wall with no history of localised pain around the swelling. Patient also complained of chest pain

predominantly on left side which was non radiating in nature for duration of 2-3 days. There was no history of hemoptysis and wheeze. There was no occupational exposure to chemicals, dust and smoke.

Personal history of the patient revealed that patient was chronic alcoholic, non smoker, there was no history of any drug addiction .Patient has no history of Type 2 diabetes mellitus and hypertension.

On examination, patient was moderately built. There was chest wall asymmetry with a chest wall swelling on left side which was soft, non tender, measuring 3-4 cm with no inflammatory signs. On auscultation of the chest left side was having diminished breadth sounds over left infrascapular area. There was no pallor, cyanosis, clubbing, pedal edema and lymphadenopathy.

Patient's serological investigations revealed anemia with Hb 10.2gm%, elevated leukocyte count with 68% polymorphs and elevated ESR of 55mm. Renal and liver function tests were within normal limits. HIV serology was non reactive.

Chest X-ray revealed blunting of the left costophrenic angle with homogenous opacity in left lower zone with thickening of the pleura on the affected side. Pleural aspiration from the left 5^{th} intercostal space was performed in which 500 cc of thick caseous pus was aspirated then post aspiration chest x-ray was done (Fig 1). Aspirated pus was sent for AFB, CBNAAT, gram staining and culture sensitivity to make out the diagnosis. Pus for AFB came out to be negative, on CBNAAT MTB sensitive to rifampicin was detected and gram staining was also negative. Sputum for AFB and CBNAAT of the patient was negative. Diagnosis of extrapulmonary tuberculosis was made and patient was started on anti tuberculosis drugs according to NTEP guidelines.

Simultaneously CECT thorax (Fig 2) was also done which revealed the surprising results. In the visualised sections of abdomen there was generalized atrophy of body and tail of the pancreas with parenchymal calcification seen in head region with main pancreatic duct dilation. Fluid collection was seen in the lesser sac extending to the left pararenal space contiguous with peri splenic cystic collections extending to the thorax via foramen of Morgagni to form large multiloculated collections in the left side also extending along the mediastinal pleura and forming large cysts in the left pericardiac and hiatal regions, which compressed the left ventricle. The bulk of the cystic collection is in the mediastinum and was associated with the extension through the left posterolateral chest wall into extra thoracic compartment forming empyema necessitans. There is compression atelectasis of the left lower lobe. Impression of chronic pancreatitis with multiloculated complicated pseudocyst formation with pancreaticopleural fistula, extensive left sided thoracic extension with empyema necessitans was mentioned. This was incidental findings which were discovered.

Due to financial constraints MRCP and ERCP of the patient could not be performed. However, patient's serum amylase and lipase levels were raised that is 354U/l and 460U/l respectively. Patient was treated with appropriate antibiotics and along with anti tubercular drugs conservatively and was put on regular follow up for further surgical procedures if needed.

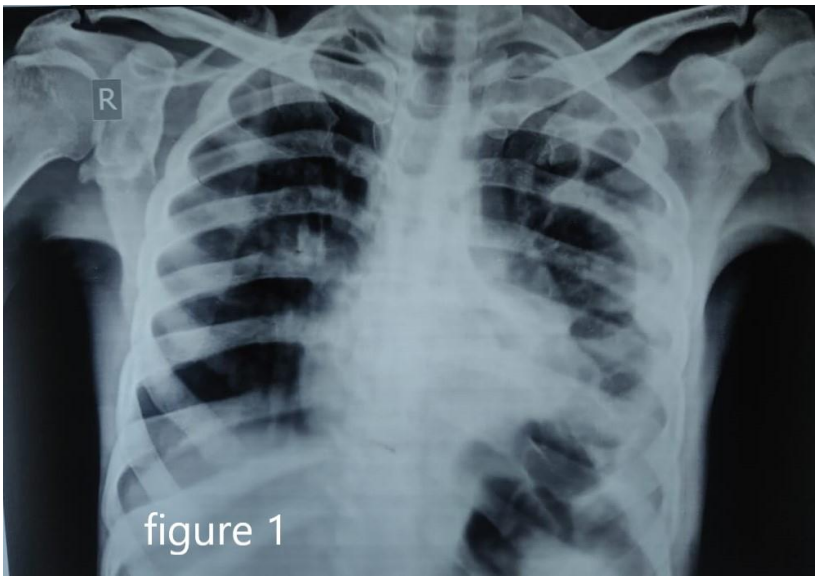
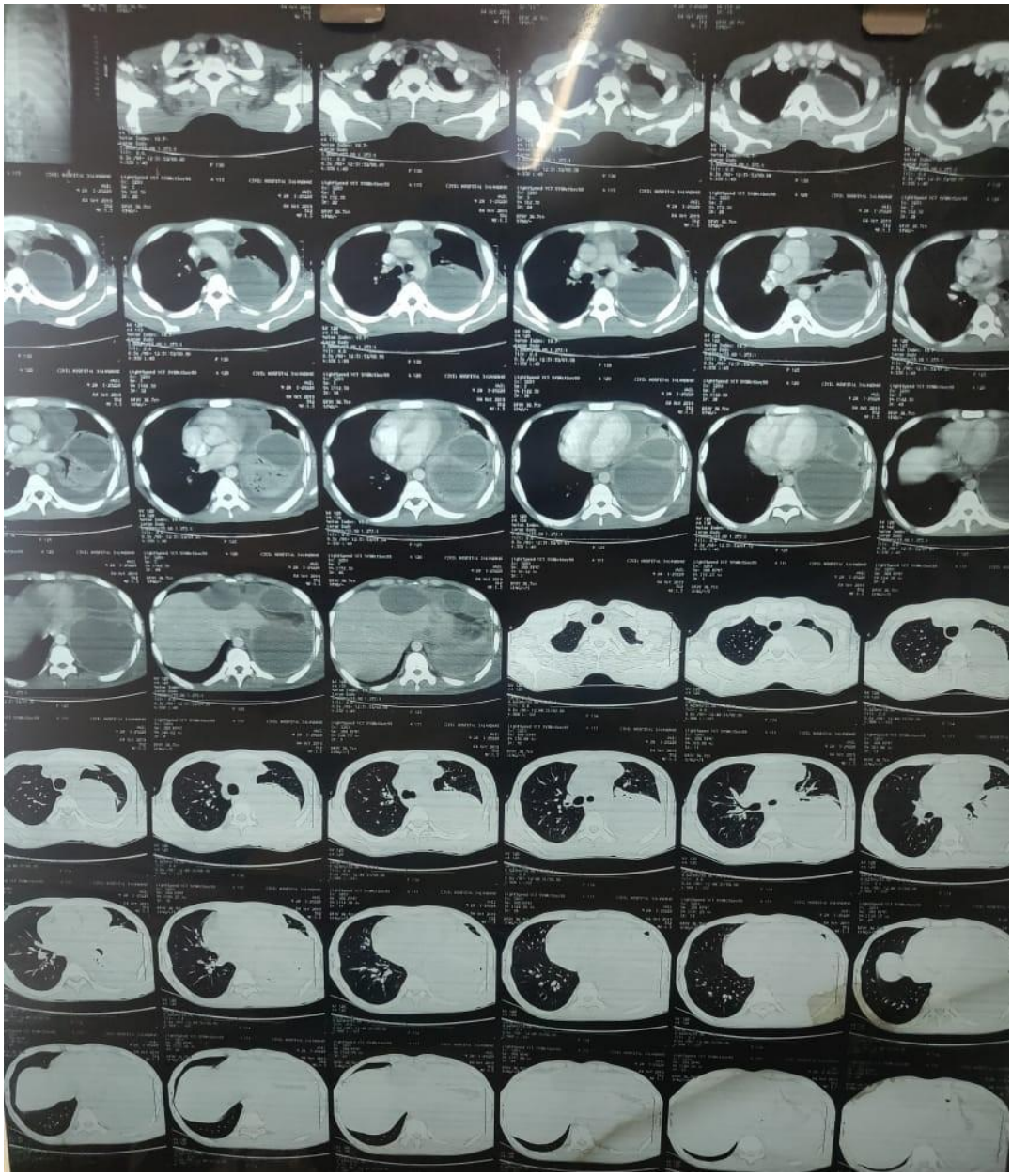


figure 1



DISCUSSION

Empyema necessitans is a rare complication especially in today's era where antibiotics are developed. The most common etiology being mycobacterium tuberculosis.[6] It can also be caused by other organisms like Streptococcus, Staphylococcus, Actinomycosis, etc. But the endemicity of a disease in the particular area in medical history plays an important role. Therefore, in the our case TB would be the first differential diagnosis. It usually develops in fibrous scar tissue resulting from pleurisy, artificial pneumothorax, or thoracoplasty.[7] Extrapulmonary TB consists of 15% of total TB. Chest wall abscesses are seen in less than 15% of musculoskeletal TB.[8,9] Tubercular pleurisy accounts for 30% to 80% of all pleural effusions in developing countries and occurs in up to 30% of patients who present with pulmonary tuberculosis.[10,11] Pleural effusion in tubercular pleurisy develop due to intra pleural hypersensitivity to mycobacterium antigens which are released due to rupture of subpleural caseous foci into the pleural space. The procoagulant effect created by the invasion of bacteria results in production of fibrin which deposits on pleural surfaces and formation of intrapleural loculations begins with formation of septas. This eventually results in pus formation and frank empyema.

Lung CT is helpful in establishing the diagnosis. The sign is connection of pleural effusion with extrapleural mass of chest wall.[12] CT scan findings are as follows: ribs damage, pleural thickening, and/or its calcification.[13] Preparation of smears for acid fast bacilli and CBNAAT is helpful. Our patient was CBNAAT positive for MTB which was rifampicin sensitive. Surgical histologic samples do have false negative results, and only 20% of cases lead to definite diagnosis.[14] Bronchopleural fistulas, pyopneumothorax, purulent pericarditis, pulmonary abscess, peritonitis from

extension through diaphragm and osteomyelitis of adjacent rib are the few complications of empyema.

Treatment can be medical as well as surgical. In the present case, anti tubercular drugs was started with adequate aspiration of pus by aspirative drain from chest wall swelling and needle aspiration from intrapleural space. Removal of all involved tissues such as bones and cartilages is an assured and safe approach but sometimes spreads the infection.[8,15] Decortication can better the lungs' function significantly.[16] The mortality rate of the surgery is 5% and is only done if there are extensive lesions reported.

Pleural effusion due to pancreaticopleural fistula is very rare as it accounts for less than 1% of cases, while pleural effusion can be seen in 3–7% of patients with pancreatitis.[17,18] Usually the diagnosis of pancreatico-pleural fistula is delayed unless there is a high index of suspicion especially in alcoholic patients or known cases of chronic pancreatitis along with pleural effusion. The delay in diagnosis usually ranged from 12 to 49 days.[19] Approximately 5% of patients with a pancreatic pseudocyst have a pleural effusion.[20] In chronic pancreatitis, the cause of pleural effusion is attributed to the direct extension of a pseudocyst across the diaphragm. or by the formation of a fistulous tract between the pancreas and pleural spaces.[21] Alcoholic pancreatitis is mostly a cause of pleural effusion as alcohol leads to acute inflammation in pancreatic duct system and elicits the protein plug formation. If transient obstruction occurs with protein plugs, pleural collection can be observed due to leakage of the pancreatic fluid. From the retroperitoneal space, it usually moves upward due to the transdiaphragmatic pressure gradient between the abdominal and pleural cavities.[22] Therefore, this explains the predominance the symptoms in men.

The single most important and reliable investigation for diagnosing pancreatico pleural fistula either due to pancreatitis or pseudocyst is determining the elevated levels of serum amylase and serum lipase. Once elevated amylase levels from a diagnostic pleural tap confirms the suspicion of a PPF, several imaging modalities are available for visualisation of the fistula. CT, ERCP and MRCP are most widely used in current practice, and the sensitivity of each modality in detecting PPF is 47%, 78% and 80%, respectively.[23,24]

Conservative treatment with pancreatic duct stenting and/ or octreotide has been successful in 31–45% of cases, while surgery is curative in 80–90% of cases but with a high rate of mortality (up to 10%).[25,26] Lymphoma, leukemia, liver cirrhosis, hydronephrosis, and pulmonary tuberculosis can also account for elevated pleural fluid levels of amylase, however, very high levels of amylase in pleural fluid can be explained only by the rupture of pancreatic pseudocyst into pleural cavity or the presence of pancreatico-pleural fistula.[27]

CONCLUSION

Tuberculosis is endemic in india and occur in association with various spectrum of diseases. This case is reported to highlight the association between extrapulmonary tuberculosis with pancreatic pathologies. As, in our patient long standing neglected pleural effusion which can be related to chronic pancreatitis and pancreatic pseudocyst leading to pancreatico pleural fistula got superadded with mycobacterium tuberculosis infection. As initially patient was asymptomatic it lead to delaying of the diagnosis resulting in empyema necessitans, the main presenting complaint. Awareness of association between the two pathologies with high index of suspicion based on clinical as well as radiological findings will help in early diagnosis subsequently reducing the morbidity and mortality.

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