

Contarini's Syndrome: A Rare Syndrome of Bilateral Pleural Effusion with Different Etiology

Sruthi Vijayan¹, N T AWAD², Jairaj P Nair³, Swapnil M Thorve⁴

ABSTRACT

Introduction: Bilateral pleural effusions usually have same characteristics and same aetiology. Contarini's syndrome refers to the occurrence of bilateral pleural fluid accumulation which can be explained by a different cause for each side. Literature search shows very few case reports with one exudative and other transudative cause like malignancy and heart failure; parapneumonic effusion with liver cirrhosis.

Case report: Here we present a rare case bilateral exudative pleural effusion, left tuberculous and right pancreatic effusion.

Conclusion: Bilateral aspiration should be performed in case of clinical suspicion of alternate diagnosis and radiological features.

Keywords: Contarini's Syndrome, Pleural Effusion, Exudative

INTRODUCTION

Pleural effusion is not an uncommon presentation for an internalist and pulmonologist. Nor is bilateral pleural effusion. Transudative pleural effusions due to cardiac, hepatic and renal disease present commonly with bilateral effusion. Usually the characteristics of pleural effusion on both sides is same and pinpoints to a single aetiology. So clinically diagnostic aspiration is done from the side of more effusion. The fluid is investigated as per differential diagnosis and treatment for the cause is initiated. Rarely pleural fluid aspiration from other side is recommended.

Contarini's syndrome is bilateral pleural effusion with different aetiology. This is uncommonly reported. Example Malignancy with cardiac failure; pneumonia with liver cirrhosis. Reported cases are usually of an exudative and transudative effusion. Here we present a rare bilateral exudative contarini's syndrome. This is a case of 40 year old male with right pancreatic and left tuberculous effusion. We haven't come across such case on search engines like pubmed and google scholar .

CASE REPORT

40 year old male, a chronic alcoholic presented with low grade fever, cough and breathlessness since 10 days. Patient was evaluated and nasal swab RT PCR for covid-19 and was detected positive. He was admitted and treated as a mild case of covid-19. Chest Xray was suggestive of bilateral pleural effusion (left more than right).

Pleural fluid was aspirated(L), which was straw yellow in appearance, lymphocytic predominant, exudative and ADA 52. Pleural fluid gene Xpert showed MTB detected with no rifampicin resistance. Patient was started on first line

antitubercular treatment and discharged.

He presented to hospital again with progressive increase in breathlessness, cough and mild abdominal pain. Patient was not having complaints of fever ,abdominal distension. He was evaluated with Chest Xray which showed increase in right sided pleural effusion. Pleural effusion on left side was decreased radiologically. USG chest was suggestive of moderate to gross right sided pleural effusion and mild left sided pleural effusion. Pleural fluid aspiration was done on right side which showed cola coloured fluid. Pleural fluid was exudative, with amylase of 1034 and lipase of 881. Pleural fluid gene xpert was negative. We subjected the patient for ultrasound guided left sided pleural fluid aspiration which was straw yellow, exudative and lymphocytic predominant. As refilling was present after therapeutic aspiration, intercostal drain was inserted on right side.

USG abdomen showed heterogenous echotexture of pancreas with evidence of 2.8*1.8 cm hypoechoic collection suggestive of pseudocyst of pancreas. CECT Chest and abdomen was done which showed gross pleural effusion on right side, collapse consolidation with mild effusion on left side, shift of mediastinum on left side, acute pancreatitis and pseudocyst of pancreas.

MRCP was done which showed multiple collections in body and tail of pancreas largest measuring 3.1*3.1*3.7 cm. collection was seen extending in to right thoracic cavity with a track of 14 mm in diameter. There was no connection between collection and left pleural cavity.

ERCP was done and selective pancreatic duct cannulation was achieved. Intercostal tube drainage was reduced to less than 50 ml and pancreatic collection was significantly decreased in a follow up ultrasound abdomen after 10 days. ICD tube was removed. Patient was discharged as was symptomatically better on antitubercular treatment.

¹Junior Resident, Department of Respiratory Medicine, LTMGH, Sion, Mumbai, ²Professor and HOD, Department of Respiratory Medicine, LTMGH, Sion, Mumbai, ³Associate Professor, Department of Respiratory Medicine, LTMGH, Sion, Mumbai, ⁴Assistant Professor, Department of Respiratory Medicine, LTMGH, Sion, Mumbai, India

Corresponding author: Dr Sruthi Vijayan, Room No 322 Old RMO Quarters, LTMGH, Sion , Mumbai 400022, India

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Figure-1: Physical appearance of left sided pleural effusion



Figure-2: physical appearance of right sided pleural effusion

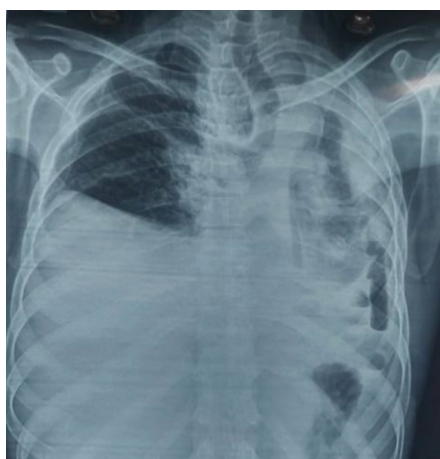


Figure-3: Chest Xray PA view- left mild pleural effusion with underlying collapse of the lung and shift of trachea to same side. Right moderate pleural effusion.

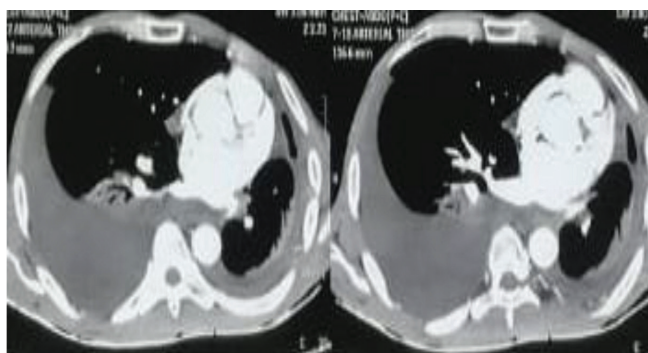


Figure-4: CECT Chest, contrast images- left mild pleural effusion with underlying collapse of the lung and shift of mediastinum to same side. Right moderate pleural effusion.

DISCUSSION

Contarini's syndrome refers to the occurrence of bilateral pleural fluid accumulation with each side due to a different cause.¹ Francesco Contarini was the 95th Doge of Venice and died in 1625. His post-mortem examination revealed that he had right hydrothorax due to Heart Failure as well as left empyema.² It is a very rare condition: its prevalence



Figure-5: CXR post ERCP- left pleural effusion with underlying collapse. Right sided mild pleural effusion with intercostal drain. ERCP stent seen in situ.

was 0.9% in a review of 546 patients with BPE and 5.6% in a prospective study of 36 patients.³

There are no clinical guidelines for bilateral pleural effusion. If the clinical diagnosis is of obvious transudative effusion we suggest of treating the cause then diagnostic aspiration. If there is diagnostic dilemma, or exudative effusion is suspected diagnostic aspiration of pleural effusion is done. In case of bilateral effusion we do aspiration from the side with more effusion.⁴ Once diagnosis is established we start on treatment for the cause of effusion suspecting same aetiology. Rarely we suspect different aetiology. We only perform aspiration from other side if the treatment fails.

Our way of approach is supported few studies. Kalomenidis et al⁵ studied 27 patients with bilateral pleural effusions who underwent bilateral thoracentesis. They found that the main biochemical and cellular features on both sides were generally similar, except for 2 (7.5%) cases which had significantly different pleural fluid lactate dehydrogenase (LDH) levels. Although a plausible explanation for the latter was not given, this circumstance did not change the categorization of the effusions. The authors concluded that bilateral diagnostic thoracenteses were not necessary unless there was a specific clinical indication.

Similarly Porcel JM et al¹, studied a database of 2605 patients. He found that only 21% had bilateral effusion and only 0.9% had bilateral effusion with different cause. In his extensive search only 12 case, 4 cases were of chylothorax and malignant effusion which explain a cause for other.

CONCLUSION

In conclusion, contarini's syndrome is very rare, not probably underdiagnosed. Though very rare bilateral aspiration should be performed in case of clinical suspicion of alternate diagnosis and radiological features on both sides like unilateral loculations, unilateral pleural enhancement or unilateral pulmonary involvement. Patient should also undergo aspiration from other side if clinical and radiological improvement is not seen after starting of treatment.

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