

Bilateral Aspiration of Pleural Fluid in a Case of Contarini Syndrome

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ABSTRACT

Contarini syndrome refers to the presence of bilateral pleural effusions with different characteristics resulting from distinct aetiologies on each side. The syndrome was named after Francesco Contarini, the 95th Doge of Venice, who had right-sided transudative effusion possibly due to heart failure, and left-sided exudative effusion due to parapneumonic effusion. While bilateral pleural effusions typically exhibit similar characteristics and are secondary to the same underlying cause, such as heart failure or malignancy, there have been rare reports of bilateral effusions with different characteristics. In this case report, we present the case of a 53-year-old female with known breast carcinoma who presented with right-sided exudative pleural effusion and left-sided transudative pleural effusion, each with distinct aetiologies, thus establishing Contarini syndrome. This report highlights the importance of considering the possibility of two separate aetiologies for bilateral pleural effusions, which should not be overlooked in clinical practice.

Keywords: Exudative, Malignant, Pleural effusion

CASE REPORT

A 53-year-old female presented to our pulmonary medicine department with complaints of breathlessness on exertion (mMRC Grade 2), chest pain lasting for two weeks, along with weakness, loss of appetite, and weight loss. She had no past medical history of diabetes mellitus, hypertension, hypothyroidism, hyperthyroidism, or seizures.

Two years ago, she was diagnosed with breast carcinoma through Fine Needle Aspiration Cytology (FNAC) from the left breast however, she refused treatment and opted for Ayurvedic medicine for the past two years.

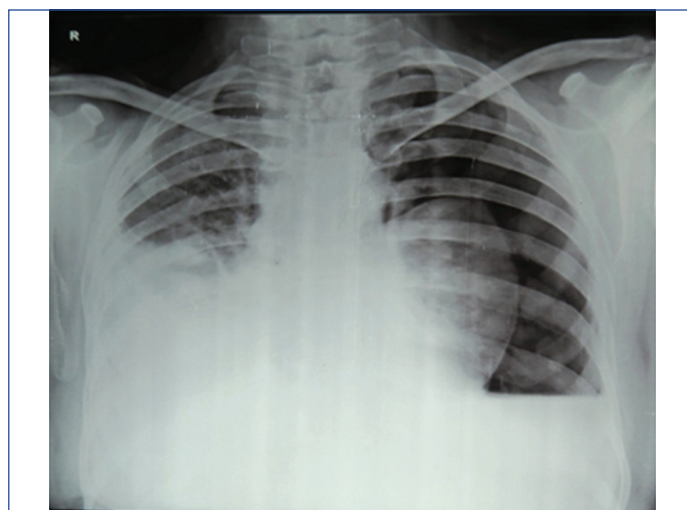
On examination, the patient was conscious, oriented to time, place, and person. Her Body Mass Index (BMI) was 18.6 kg/m². She appeared anaemic and emaciated, with tachypnoea (respiratory rate of 30 breaths/min). All other vitals were normal. Physical findings included pallor, bilateral pitting pedal oedema, and enlarged axillary lymph nodes. Auscultation revealed decreased breath sounds bilaterally in the interscapular and subscapular regions. The cardiovascular, genitourinary, gastrointestinal, and central nervous systems showed no abnormalities upon clinical examination.

Physical examination of the left breast revealed a hard, erythematous, ulcerating mass lesion measuring approximately 7×5 cm in the subareolar region, accompanied by palpable enlarged axillary lymph nodes. The right breast and axilla appeared normal.

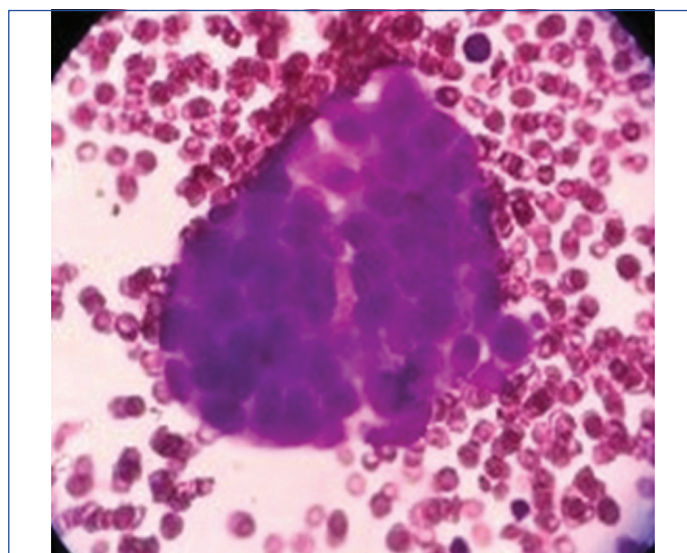
Chest X-ray (PA view) exhibited right-sided pleural effusion and left sided hydropneumothorax [Table/Fig-1]. Haematological tests, apart from a haemoglobin level of 6.5 g/dL, were inconclusive. Ultrasound Sonography Test (USG) thorax revealed a notable disparity in the fluid level between both pleural spaces.

The total serum proteins and differential serum proteins were found to be on the lower side, measuring 4.2 g/dL, with albumin levels at 1.6 g/dL and globulin levels at 2.6 g/dL, indicating hypoalbuminaemia in combination with hypoproteinaemia. Pleural fluid aspiration was performed on the left side, revealing haemorrhagic fluid. The pleural fluid analysis indicated an exudative nature (pleural fluid protein/serum protein ratio of 0.7), as per the Light's criteria [1]. The pleural fluid showed an Adenosine Deaminase (ADA) level of 10 IU/L, with a predominance of lymphocytes and positive cytology for atypical cells (suggestive of malignancy) [Table/Fig-2]. The Cartridge-Based

Nucleic Acid Amplification Test (CBNAAT) assay performed on the pleural fluid yielded a negative result.

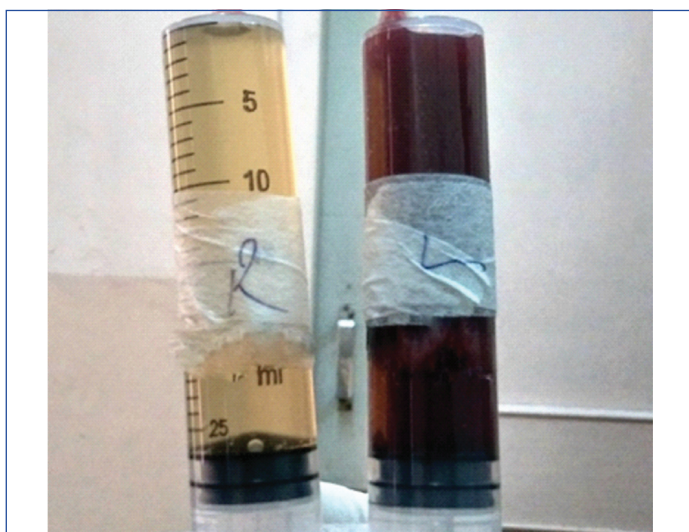


[Table/Fig-1]: A chest X-ray (PA view) showing right-sided pleural effusion and left-sided hydropneumothorax.



[Table/Fig-2]: Cytology showing atypical cells (40x) with May Grunwald-Giemsa stain.

A tube thoracostomy was performed on the left side, and suspecting bilateral malignant pleural effusion, diagnostic aspiration was also done on the right side. Clear, watery, slightly yellowish fluid was obtained from the right side [Table/Fig-3]. The fluid was found to be transudative (pleural fluid protein/serum protein ratio of 0.02) according to the Light's criteria. Pleural fluid pH was not checked, but pleural fluid LDH was measured at 140 IU/L, and leucocyte count was 800/mm³. Clinical Value of Contrast-Enhanced Computed Tomography (CECT) of the abdomen did not reveal any abnormalities, and echocardiography showed normal left ventricle function with an ejection fraction of 54%, ruling out cardiac aetiology. Liver function tests, renal function tests, and thyroid profile were within normal limits, ruling out these as the cause of the transudative effusion. Interferon-Gamma (IFN- γ) release assay and CBNAAT were negative, ruling out tuberculosis. Repeated cytological examination of the fluid from the right side showed no malignant cells.



[Table/Fig-3]: Gross appearance of pleural fluid aspirate (gross appearance) from both the right and left-sides.

The patient was diagnosed with Contarini syndrome, as she had bilateral pleural effusion due to two different aetiologies (hypoalbuminaemia on the right side and malignant pleural effusion on the left side). Pleurodesis was performed on the left side, and the patient was placed on a high protein diet and received albumin infusion to improve her overall condition. Following pleurodesis, the left-sided effusion significantly reduced, and the right-sided effusion slightly decreased. The patient experienced symptomatic improvement and was discharged. She remained on regular follow-up visits for four months before eventually passing away due to pulmonary insufficiency and respiratory failure.

DISCUSSION

Congestive heart failure, malignancy, infection, and pulmonary embolism are responsible for over 90% of pleural effusions [2]. Unilateral or bilateral pleural effusions are commonly encountered in medical practice, with bilateral pleural effusions occurring in approximately 15% of 642 patients [3]. Typically, bilateral pleural effusions are caused by a single factor, often heart failure or malignant diseases [4]. However, Contarini syndrome is an exception characterised by bilateral pleural effusions of different aetiologies with distinct pleural fluid characteristics [5]. It is not uncommon for a parapneumonic effusion to trigger heart failure, which subsequently leads to the development of a contralateral transudate [6].

Based on medical records and literature review by Porcel JM et al., only 12 cases of Contarini syndrome had been reported worldwide until 2012 [7]. Among these cases, three were associated with heart failure/empyema, four with malignancy/chylothorax, two with malignancy/empyema, and one each with pericardial disease/

simple parapneumonic effusion, subphrenic abscess/hypervolaemia due to excessive perioperative volume load, and complicated parapneumonic effusion/radiation pleuritis (total of 12 cases). The first reported case of Contarini syndrome from India was in 2004, involving a 23-year-old male smoker with Human Immunodeficiency Virus (HIV) seropositivity. The patient presented with right-sided staphylococcal empyema and left-sided clear lymphocytic predominant tubercular effusion [8]. In this case report, the patient had a malignant pleural effusion on the left side, resulting from metastasis of breast carcinoma. The transudative effusion on the right side could be attributed to lymphatic obstruction by malignant cells (paramalignant effusion), her overall poor general condition (hypoalbuminaemia in combination with hypoproteinaemia), or a combination of both processes. It is worth noting that hypoalbuminaemia can also lead to transudative pleural effusion [9].

Traditionally, it has been advised to perform thoracentesis on one side at a time when bilateral effusions are present, likely due to concerns of causing bilateral pneumothoraces or other complications [10]. However, this practice may have contributed to an underdiagnosis of this rarely reported entity, as most patients with bilateral pleural effusions undergo unilateral thoracentesis. In this case, bilateral tapping was performed to determine the exact cause of the effusions on both sides due to the following reasons:

1. Disproportionate accumulation of pleural fluid on both sides.
2. Presence of both hypoalbuminaemia and malignancy in the patient.

Contarini syndrome serves as an excellent example of 'Hickam's dictum,' which states that 'patients can have as many diseases as they damn well please.' Therefore, there is a compelling need to aspirate fluid from both sides [9].

Ferreiro L et al. recommended bilateral thoracentesis in the following situations [11]:

- 1) Presence of atypical clinical findings (such as fever or chest pain associated with decompensated heart failure).
- 2) Unilateral pulmonary parenchymal involvement.
- 3) Significant difference in size between the pleural effusions.
- 4) Markedly different attenuation values of the pleural effusion on Computed Tomography (CT) scan (Hounsfield units).
- 5) Resolution of a unilateral pleural effusion.

Therefore, the rationale for considering bilateral diagnostic thoracentesis is to avoid missing significant pathology in cases where different pathologies are present on each side, as in the indexed case. The debate surrounding whether to aspirate fluid from one side or both sides is ongoing, and the decision should be made in the best interest of the patient to reach a final diagnosis.

CONCLUSION(S)

Contarini syndrome, being a very rare entity, serves as a reminder to be vigilant when investigating cases with bilateral pleural effusions. It is important not to assume that all bilateral pleural effusions have the same appearance and aetiology. Instead, clinicians should consider the possibility of Contarini syndrome in appropriate clinical situations.

In conclusion, Contarini syndrome highlights the importance of careful evaluation and consideration of multiple aetiologies when managing patients with bilateral pleural effusions. It serves as a valuable reminder that not all bilateral pleural effusions are the same, and a comprehensive approach is necessary for accurate diagnosis and appropriate treatment.

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