Case Report

An unusual case of loculated malignant pleural effusion mimicking an infected pericardial cyst

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ABSTRACT

Malignant pleural effusion (MPE) is one of the frequent and distressing conditions identified at the advanced stage of different malignant conditions. This problem seems to affect patients with lung and breast malignancies which contribute to the lower life quality. Less commonly MPE presents as a loculated fluid collection. We present an unusual case of cystic fluid collection in contact with the heart appearing similar to a pericardial cyst, but on cytological examination turned out to be loculated MPE. Furthermore, there was the presence of malignant fissural effusion. This text highlights the unusual location of this loculated MPE along with a few characteristic features to differentiate it from the benign pericardial cyst.

Key words: Loculated malignant pleural effusion, Lung cancer, Malignant fissural effusion, Pericardial cyst

alignant pleural effusion (MPE) is defined as the build-up of a considerable amount of exudate within the pleural space, along with the presence of malignant cells or tumor tissue [1]. MPE is an important complication in patients with malignancies [2]. Management approaches to MPE cases are dependent on the age, histopathology of the primary tumor, response to chemotherapy, and life expectancy. In long term, the obliteration of the pleural cavity to stop the reaccumulation of pleural fluid should be the primary aim [3].

Pericardial cysts are uncommon cystic masses in the mediastinum. Most of them are a result of developmental abnormalities. Infrequent causes comprise echinococcosis, benign teratoma, cavernous hemangioma, and trauma. Diagnosis can be established using computed tomography (CT), transthoracic and transesophageal echocardiography, or magnetic resonance imaging. Conventional management of pericardial cyst consists of thoracotomy or, in some patients, percutaneous aspiration, along with ethanol sclerosis. Lately, video-assisted thoracoscopic surgery has been utilized with success in the management of pericardial cysts [4].

We present a case of loculated MPE along the mediastinal pleura on the left side which was masquerading as an infected pericardial cyst. To the best of our knowledge, this is the only case with such an unusual presentation of MPE as a loculated fluid collection along the mediastinal pleura with coexisting malignant fissural effusion.

Access this article online	
Received - 17 March 2022 Initial Review - 05 April 2022 Accepted - 26 April 2022	Quick Response code
DOI: 10.32677/ijcr.v8i5.3387	

CASE REPORT

A 55-year-old male patient presented with 2 months history of breathlessness on exertion, fever, and left-sided chest pain. Fever was low grade and intermittent for 2 months with no specific predilection for the time of the day. He denied any history of weight loss, loss of appetite, cough, or hemoptysis. He had no comorbidities except for hypertension diagnosed 2 years back, for which he was taking 5 mg of amlodipine once a day. He was diagnosed with sputum-positive pulmonary tuberculosis 5 years back, for which he had taken antitubercular treatment for 6 months. He had a 30 pack/years history of cigarette smoking.

On examination, blood pressure was 130/80 mmHg, pulse 82/min, body temperature was 37.1°C, and respiratory rate was 16/min. The general examination was within normal limits.

An electrocardiogram was done and turned out to be unremarkable. On respiratory system examination, there was decreased air entry in the left lower lobe. The rest of the systemic examination was within normal limits. His total leucocyte count was 20,000/cu. mm. The rest of the laboratory tests, including hemoglobin, liver function tests, and renal function tests, were within normal limits.

The patient underwent contrast-enhanced CT (CECT) of the thorax which showed a well-defined cystic lesion measuring 110 mm in the largest dimension located within the middle mediastinum in the left hemithorax in close contact with the heart with peripheral wall enhancement (Fig. 1a). A subtle fluid level was also present within the cystic cavity which was visible on

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proper windowing (Fig. 2a). There was no evidence of enhancing solid component noted within this cystic lesion favoring the possibility of a benign pericardial cyst which could have got infected due to the settled debris within the cyst along with the presence of fever and raised leucocyte count. Another loculated fluid collection with bulging contours was noted in the left oblique fissure. It showed no enhancement in the post-contrast study representing fissural effusion (Fig. 1b). Few calcified mediastinal lymph nodes were present likely secondary to the past history of tuberculosis. An enhancing soft-tissue attenuation area was noted in the inferior lingular segment of the left upper lobe which was abutting both the fluid collections and was interpreted as an area of consolidation (Fig. 2b).

CT-guided aspiration of both the fluid collections was performed (Fig. 3). The aspirated fluid from the medial collection was brownish in color and the fluid aspirated from the fissural effusion was straw-colored. On cytological examination, malignant cells were found in both the aspirated fluid samples (Fig. 4). Subsequently, the heterogeneously enhancing area in the inferior lingual segment, which was originally suspected to be a consolidation, was biopsied and turned out to be an adenocarcinoma of the lung.

The patient was started on concomitant chemoradiotherapy. The aim of the association of chemo and radiation therapy is to improve disease control at both the local and systemic levels. Because platinum-based dual chemotherapy combined with concomitant chemoradiotherapy is the optimal treatment for certain patients with the early or locally advanced malignancies, the patient was started on vinorelbine-cisplatin. A weekly

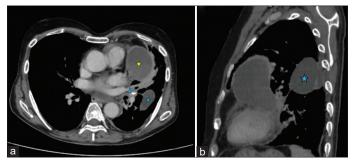


Figure 1: (a and b) Axial and sagittal sections of contrast-enhanced computed tomography thorax showing peripherally enhancing cystic lesion abutting the left heart border (yellow star). Loculated fluid collection in the left oblique fissure without post-contrast enhancement (blue star). Calcified left hilar lymph node (blue arrowhead)

external beam radiation treatment was part of the radiation therapy schedule. One month after the start of therapy, the patient developed radiation esophagitis. Apart from that, there were no other complications and the patient's symptoms have improved over time.

DISCUSSION

Pleural effusion may be the initial indication of malignancy, which usually suggests recurrence or progressed disease. The primary tumors or metastasis can permeate the visceral pleural layer which affects the usual resorption of fluid from the parietal to the visceral pleura or results in more capillary leaking and elevated fluid production.

Lung malignancies, including malignant pleural mesothelioma, are the most frequent source of MPE. In females, breast cancer remains the most common source of MPE [5]. The presence of cells with nuclear atypia in the pleural fluid indicates the presence of disseminated or advanced malignancy, which is generally linked with a shorter life expectancy [6].

Ultrasonography and/or CT are helpful in establishing the diagnosis of locules and subsequent therapeutic intervention [5]. CECT thorax is the current gold-standard imaging technique for the evaluation of pleura. Cytology is an established initial diagnostic test with a mean sensitivity of 60% with high sensitivity for adenocarcinoma (79%) and lower sensitivity for mesothelioma (6%) [1,6]. Immunohistochemistry aids in distinguishing reactive mesothelial cells from those of malignant pleural mesothelioma and adenocarcinoma metastases by exposing them to different panels of antibodies since each of them has specific antigens [6].

Pericardial cysts are infrequent, benign abnormalities present since birth and have an incidence of around 1 in 100,000 which constitute about 7% of all mediastinal lesions. They are, generally, found in the age group of 30-40 years and are more commonly found in men with a male-to-female ratio of 3:2. These are mostly situated at the right costophrenic angle (70%), then the left costophrenic angle (22%). Pericardial cysts are seen as thin-walled, uniform, well-delineated, and unenhancing cystic lesions with no internal septations. They are generally having an oval shape, but have also been described with a pointing shape or teardrop morphology and do not show enhancement with contrast administration [7].

The traditional treatment for symptomatic MPE entails either pleurodesis (which has a failure rate of 30%) or recurrent

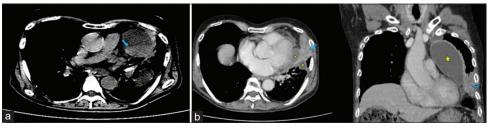


Figure 2: (a) Non-contrast axial computed tomography (CT) thorax on appropriate windowing showing settled hyperdense content within the cystic cavity representing settled debris (blue arrow); (b) Axial and coronal sections of contrast-enhanced CT thorax demonstrating a heterogeneously enhancing lesion in inferior lingular segment (blue arrow) in contact with both the loculated fluid collections (yellow stars)

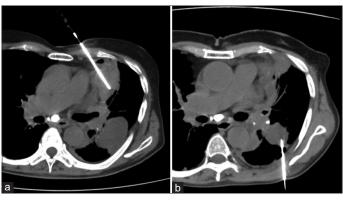


Figure 3: Axial computed tomography (CT) images showing aspiration of both the loculated fluid collections under CT guidance

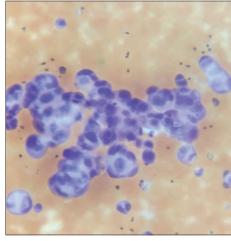


Figure 4: A smear from fluid shows sheets of cells, also cells in clusters with binucleate cells and nuclear pleomorphism on hemorrhagic background suggesting malignant cells

thoracentesis, particularly in patients with trapped lungs. Recent landmark randomized controlled studies have shown the benefits of using an indwelling pleural catheter as the first-line treatment for MPE [8]. Immunotherapy also provides a significant overall survival improvement in patients with advanced lung carcinoma. Pembrolizumab or atezolizumab as monotherapy outperforms first-line chemotherapy in malignancies with high PD-L1 expression. A lot of chemoimmunotherapy combinations outperform chemotherapy as a combined treatment [9].

It is very unusual for an MPE to present as a loculated fluid collection along the mediastinal pleura closely abutting the left heart border. Furthermore, the clinical presentation of the patient was also unusual without any typical symptoms of malignancy such as hemoptysis, weight loss, and anorexia. Pericardial cysts can infrequently be detected in the older age group as they are generally asymptomatic and usually do not show enhancement and contain clear fluid.

CONCLUSION

The presence of peripheral enhancement with slightly hyperdense cyst contents occasionally with fluid-fluid level should point toward an alternative diagnosis than a pericardial cyst, and hence, aspiration of such fluid collections for cytology is important. In presence of such atypical features in cystic fluid collection close to the heart, a heterogeneously enhancing area that might mimic consolidation should also be interpreted with caution as it could be a possible malignant lesion in the lung.

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Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Nimje VV, Bodhankar SB, Yadav TS. An unusual case of loculated malignant pleural effusion mimicking an infected pericardial cyst. Indian J Case Reports. 2022;8(5):131-133.