

Bilateral Hydropneumothorax in a patient with Metastatic Gastric Adenocarcinoma of Signet Ring Morphology- An Unusual Presentation

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ABSTRACT

Pleura is a commonest location for metastasis of malignancy of various organs. The infiltration of pleura usually present as Pleural Effusion, which is the hallmark of pleural metastasis, other presentation include pleural nodules or extensive pleural thickening and rarely as pneumothorax or Hydro-pneumothorax as noted in this case report. Lung carcinoma is the most frequent malignancy that invades pleura thereby resulting in malignant and paramalignant effusions. Carcinoma of Breast is second frequent malignancy leading to malignant pleural effusion.

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Introduction

Pleura is a commonest location for metastasis of malignancy of various organs. The infiltration of pleura usually present as Pleural Effusion, which is the hallmark of pleural metastasis,¹ other presentation include pleural nodules or extensive pleural thickening and rarely as pneumothorax or Hydro-pneumothorax as noted in this case report. Lung carcinoma is the most frequent malignancy that invades pleura thereby resulting in malignant and paramalignant effusions. Carcinoma of Breast is second frequent malignancy leading to malignant pleural effusion. Altogether about 65% of all malignant pleural effusions accounts for these two malignancies while ovarian and gastric carcinoma representing about 5% of pleural effusion². About 10% cases of all malignant pleural effusion occur in

Lymphoma.² Signet-ring cell carcinoma of the stomach is seen in younger age predominantly in female and has a fulminant and rapidly progressive course.³ Here we are reporting a case of mucin-secreting signet-ring cell Adenocarcinoma of stomach with metastatic bilateral hydropneumothorax which is an unusual phenomenon. To the extent of our knowledge, this is the first report of bilateral Hydropneumo-thorax associated with metastatic Adenocarcinoma with primary in stomach.

Case Report

A 25-year-old housewife, nonsmoker, presented with 2 months history of Productive cough, decrease appetite and shortness of breath which worsens for 4 to 5 days prior to hospital admission. Six months back she had undergone Total abdominal hysterectomy

and bilateral Salpingo-oophorectomy after workup for abdominal pain and distension. Biopsy of omentum, right ovary, right fallopian tube and left ovary exhibit

Metastatic Adenocarcinoma of stomach with Signet Ring morphology. Tumor Markers CA-125 was raised 60.25 (reference range: 0-35 U/L). Cytokeratin 7,

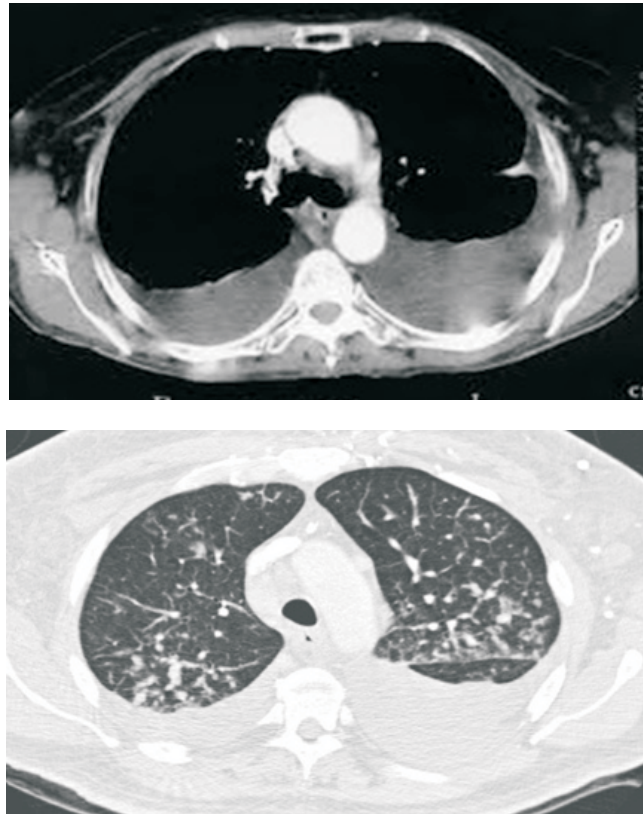


Fig. 1.1: Contrast Enhanced CT Chest Showing Bilateral Pleural Effusion

cytokeratin 20 & CDX-2 were also positive. Based on the biopsy report, chemotherapy was commenced with Oxaliplatin and Capecitabine and 4 cycles of chemotherapy were given. CT scan shows multiple small lymph nodes in the neck at various levels, left jugular vein & left brachiocephalic vein were distended with dense lumen appearance likely due to deep venous thrombosis. Subcutaneous swelling and edema noted in left axillary region and lower neck, multiple lymph nodes in mediastinum, mesentery, retroperitoneal region, mild bilateral pleural effusion (Fig. 1.1) and lytic lesion in L2 vertebrae. Deep venous thrombosis of left jugular vein and left brachiocephalic vein was treated accordingly.

A follow up chest CT scan showed progression of the disease with moderate bilateral pleural effusion. Multiple lymph nodes in mediastinum, abdominal & retroperitoneal region. Lytic lesion in L2 & L4 vertebrae. Consequently, patient was referred to Ojha Institute of chest diseases for specialist evaluation and further management of Pleural effusion.

On Admission, patient was dyspneic, hypotensive and tachypneic, not maintaining oxygen saturation at room air, immediately after resuscitation chest X-ray was done which showed Bilateral lamellar pleural Effusion (Fig. 1.2 A) for which chest drains were inserted on right side (Fig. 1.2 B). After drainage of hemorrhagic fluid the subsequent Chest radiograph showed an air-fluid level with bubbling of air in under water seal (Fig. 1.3). Sputum studies (AFB smear & MTB G-Xpert) & routine culture was found to be negative. Pleural Fluid Studies showed exudative lymphocytic picture. Pleural fluid AFB smear was negative and MTB on GeneXpert was not detected. Pleural fluid culture was negative. Pleural Fluid cytology showed malignant cells. There was drainage of about 800ml of fluid in chest drain daily. Patient became dyspneic again after 3 days, so thoracostomy was carried out on left side. About 5400ml of fluid drained from right side of chest & 4600ml from left side. Follow up chest radiograph showed Hydropneumothorax on both sides (Fig. 1.4A, B) with hemorrhagic fluid drained from both sides and

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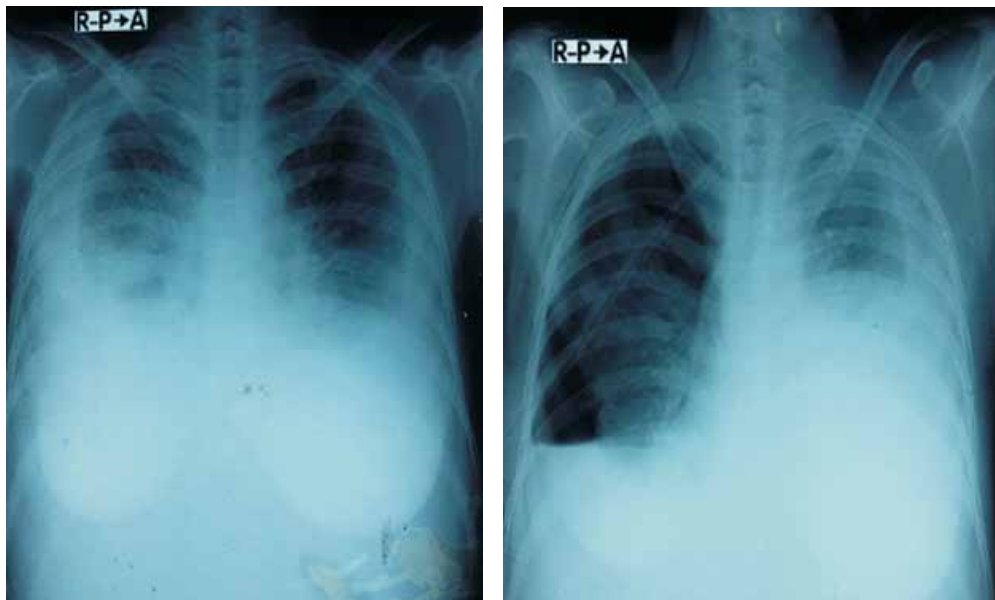


Fig. 1.2: Chest X-ray:

A; Bilateral Pleural Effusion-at time of admission (Right)

B; Rt. Sided Hydropneumothorax (Left)



Fig. 1.3: Chest Drain showing Hemorrhagic fluid

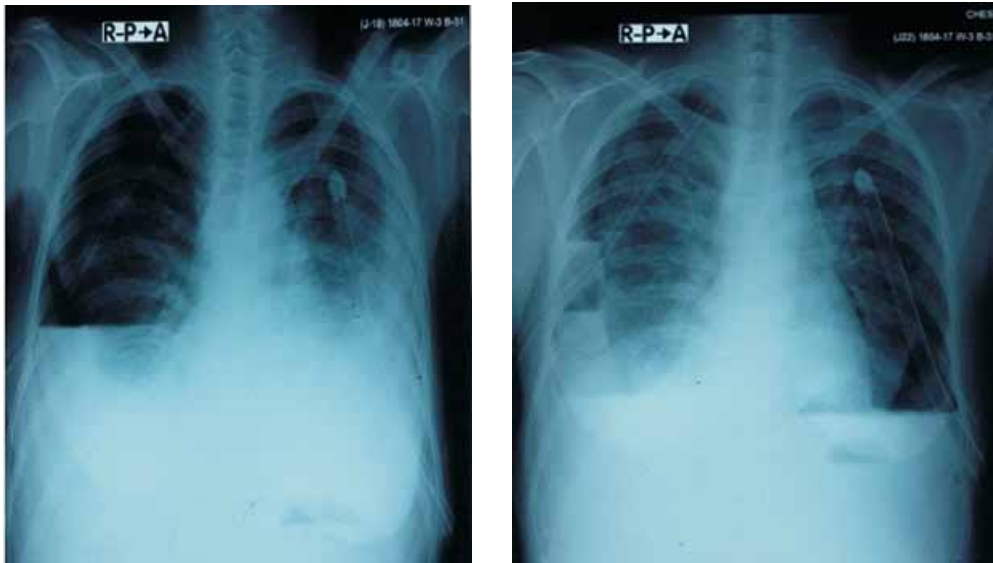


Fig 1.4: Chest X-ray

A; Hydropneumothorax (Right)

B; After 4 days of Tube thoracostomy- Bilateral hydropneumothoraces (Left)

Her clinical symptoms improved after pleural fluid drainage. After few days, she developed frank hematuria. Ultrasound Whole Abdomen showed mild ascites and left Hydronephrosis with possible metastasis in left kidney. On day 7 of admission patient expired due to disease progression.

Discussion

Pleura is a commonest location for metastasis from distant organs. Metastasis most often manifest as pleural masses, single or multiple nodules, pleural effusion & rarely pneumothorax.¹ Hematogenous spread, lymphatics, or direct contact by malignant neoplasms are the main mechanism of pleural metastasis.⁴

Pleural metastasis and malignant effusion occurs with a variety of cancers. Characteristic feature of the parietal pleura is lymphatic stomata which is the openings between parietal pleural mesothelial cells forming lymphatic lacunae just below the mesothelial layer.⁵ These lacunae consolidate into collecting lymphatics, then into intercostal trunk vessels. The direction of flow is mainly toward the mediastinal lymph nodes.⁵ The most important function of lymphatics of parietal pleura is absorption of pleural liquid and proteins. Hence, Pleural effusion results when there is interference with the integrity of the lymphatics anywhere between parietal pleura and mediastinal lymph nodes.⁵

Patient with Carcinoma involving pleura mostly present with symptoms attributable to a large Pleural Effusion, Dyspnea on exertion and Cough. Detection of exfoliated malignant cells in pleural fluid or presence of malignant cells in pleural tissue taken from percutaneous pleural biopsy, thoracoscopy, thoracotomy, or at autopsy is required for diagnosis of malignant pleural effusion.² In majority of patients, pleural effusion results from malignancy but neoplastic cells cannot be detected in pleural fluid or pleural tissue. So such pleural effusions associated with malignancy, in which no direct pleural involvement with tumor and no other cause for effusion is labelled as paramalignant effusion.² Paramalignant pleural effusion occurs from local effects of tumor like lymphatic obstruction, bronchial obstruction also systemic effects such as pulmonary embolism & hypoalbuminemia as well as adverse effects of therapy.

Spontaneous pneumothorax in metastatic disease is rare and mostly manifest in advanced disease or during cytotoxic therapy.⁶ Theories regarding possible mechanisms proposed that Spontaneous rupture of necrotic tumor tissue or of oncological treatment may create bronchopleural fistulae. Secondly, intermittent nodular bronchiolar obstruction at the lung periphery may lead to subpleural blebs.⁶ It is thought that probably tumor invasion cause disruption of the visceral pleura and of peripheral bronchioles might produce persistent slow air

leakage resulting in small and clinically silent pneumothorax.⁶ Though in our case there was no involvement of lung apparent on CT scan. A case of metastatic Synovial sarcoma reported from Qatar showed bilateral pneumothorax during chemotherapy.⁶ Hydropneumothorax presenting as metastasis of malignancy is extremely rare. In a rare presentation in two case reports showed Recurrent or simultaneous Hydro-pneumothorax secondary to malignant mesothelioma which presents nonspecifically and in this case uniquely as a Hydro-pneumothorax. The mechanism of Hydro- pneumothorax is unclear. It is thought to result from rupture of necrotic tumor nodules.⁷⁻⁹

Worldwide, Gastric cancer is fourth frequent malignancy, after cancers of the lung, breast, and colon & rectum.¹⁰ According to World Health Organization, in

2012 stomach cancer accounts for 723,000 deaths worldwide.¹⁰ Gastric cancer is a major public health problem, approximately 951,000 new cases diagnosed over the globe in 2012, accounting for 6.8% of all new cases of cancers. According to the WHO's classification Gastric Signet ring cell carcinoma is a poorly cohesive carcinoma comprising of tumor cells with prominent cytoplasmic mucin and a crescent-shaped nucleus eccentrically placed (Fig. 1.4).¹¹ Gastric Signet-ring cell carcinoma tends to spread transversely in early gastric cancer but once it invades the submucosal region, it shows high potential to metastasize.¹² The signet ring cell histologic type is also known to be linked with advanced stage. In another reported case, an unusual presentation of Signet cell Carcinoma was with massive pulmonary tumor embolism in young patients.^{13,14}

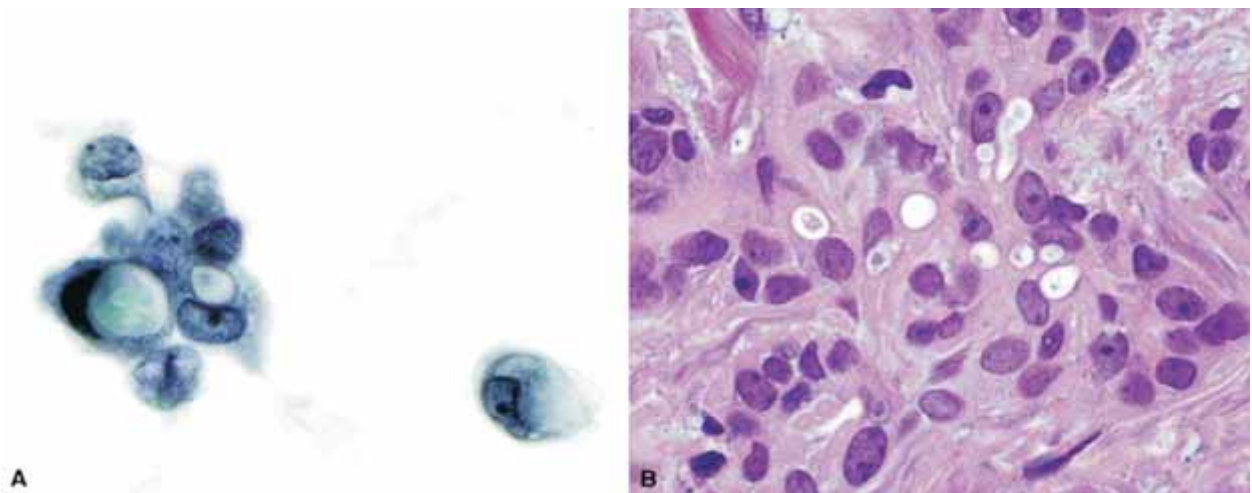


Fig. 1.4: A, Metastatic adenocarcinoma in a fine-needle aspiration specimen showing focal “signet ring cell” differentiation with formation of cytoplasmic vacuoles. B, The same feature is seen in this cell block preparation.

An awareness of the spectrum of radiologic appearance in atypical pulmonary and pleural metastases along with histopathologic correlation allows the distinction of metastases from other nonmalignant diseases. Treatment of such Hydropneumothorax related to metastasis may be difficult especially if the underlying mechanism is Broncho-pleural fistula which could lead to persistent air leak in spite of chest tube insertion which will mandate surgical intervention or chemical pleurodesis from the initial presentation.

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