Case report

Recurrent hydropneumothorax: An unusual presentation for malignant pleural mesothelioma

David DeLapp a, *, Christopher Chan a, Perry Nystrom b

a Wright State University Boonshoft School of Medicine Internal Medicine Program, Miami Valley Hospital, Weber CHE Building, 128 E. Apple St., 2nd Floor, Dayton, OH, 45409, USA
b Dayton VA Medical Center, 4100 W. Third St., Dayton, OH, 45428, USA

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A B S T R A C T

Mesothelioma is a rare pulmonary malignancy commonly associated with asbestos exposure. Its presentation is insidious and non-specific, with complaints of chest pain, dyspnea and cough. Chest X-ray may demonstrate unilateral pleural effusion. CT and PET scans may highlight nodular pleural plaques. Diagnosis often times is difficult with negative imaging and negative pleural fluid studies. In rare cases, hydropneumothoraces may be seen. We report a case of malignant pleural mesothelioma presenting as recurrent hydropneumothorax with negative CT scan of the chest for pleural abnormalities and negative pleural fluid studies.

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1. Introduction

Mesothelioma is a rare (1% of all cancers) and lethal disease with a median survival of 9–17 months. It is most commonly associated with asbestos exposure [1,2]. Currently, there are about 2500 cases per year in the US. While the incidence has stabilized, it is on the rise in other countries [3]. It is typically diagnosed after patients present with nonspecific symptoms of chest pain, dyspnea, and cough with initial imaging most commonly showing unilateral pleural effusion. The combination of nonspecific symptoms and imaging findings can make mesothelioma a diagnostic dilemma. This case highlights the unusual ways in which mesothelioma can present, and the importance of a thorough history taking and the necessity for further investigation with recurrence of hydropneumothoraces or unilateral pleural effusions.

2. Case

A 67 year-old male with a history of hypertension and hyperlipidemia presented to his primary care provider with dyspnea and productive cough for two weeks. He denied weight loss, fevers, or hemoptysis. He was a former cigarette smoker who served four years in the Navy about 40 years ago. Vitals signs and physical exam, including breath sounds, were normal. Initial chest X-ray revealed a right hydropneumothorax. Right tube thoracostomy and doxycycline pleurodesis were performed during a short hospital admission. Approximately 1.5 L of exudative pleural fluid was removed, but cytology was negative for malignant cells. He returned eight months later with progressive dyspnea, orthopnea, and cough. Chest X-ray and CT of the thorax showed a large right pleural effusion without pleural abnormalities (Fig. 1). A diagnostic and therapeutic right-sided thoracentesis was notable for exudative pleural fluid which was again unremarkable for malignant cells. A PET/CT scan was negative. He returned to the hospital two more times over the next two weeks for dyspnea and found to have a right-sided pleural effusion on CT scan of thorax (Fig. 2) followed by recurrence of right hydropneumothorax on chest X-ray (Fig. 3). Further investigation included a thoracoscopy and pleural biopsy, which demonstrated a diffuse micronodular and visceral pleura. Pathology showed epithelioid mesothelioma that was involving the visceral pleura and was staged T2 N2 M0. Subsequently, he had mediastinoscopy, extrapleural pneumonectomy, right pericardiectomy, and right hemidiaphragm removal and reconstruction followed by chemoradiation. The patient did well for about 1 year until he returned with a right hydrothorax and passed away a few months later.

3. Discussion

This case highlights the difficulties in diagnosing malignant
Mesothelioma due to nonspecific symptoms and non-diagnostic imaging studies. Not only did the patient present with nonspecific symptoms, but the initial CT scan did not show any pleural abnormality, and pleural fluid analysis was negative for malignant cells. Few case reports have noted recurrent hydro pneumothorax as the only presenting feature of mesothelioma [4–9].

Patients with mesothelioma typically present with a history of asbestosis exposure, which highlights the importance of a thorough history, as greater than 90% of cases have an association with asbestosis [1]. There is typically a latency period of 15–60 years from exposure, and patients are typically diagnosed at ages 50–70 years old [10]. Patients typically present with symptoms of chest pain and/or dyspnea that develop over time. Physical exam is nonspecific with signs of a pleural effusion being the only contributing finding. Pleural fluid is only positive in 30% of cases [1]. It is fairly common for mesothelioma to present with a pleural effusion as up to 80% of patients present with a unilateral pleural effusion [2,11,12]. Spontaneous primary pneumothoraces are also fairly common with an incidence of 18–28/100,000 men; however only 0.02% are associated with malignancy [13]. In a small case series of mesothelioma patients, 11% of patients presented with pneumothoraces [14]. There is little to no data on hydro pneumothoraces as the presenting feature of malignant mesothelioma. The mechanism of hydro pneumothorax is unclear. One popular theory is that it is caused by the rupture of necrotic tumor nodules [15].

Other imaging modalities can be more revealing, but no single imaging diagnostic tool should be used in patients with risk factors and high suspicion. Chest X-rays can show pleural masses or thickening in 45–60% of cases [16]. Pleural changes on CT chest have been noted to have up to 90%–95% sensitivity [12,17–19]. PET scans have been shown to have a 96.8% sensitivity and 88.5% specificity for distinguishing malignant disease and are typically only used for staging [20]. There were no easily notable pleural features initially on imaging in our patient despite the high sensitivity of the CT and PET/CT scan making this case even more unusual.

Mesothelioma presents nonspecifically and in this case uniquely as a hydro pneumothorax making diagnosis difficult. The case demonstrates the importance of early invasive testing with recurrence of hydro pneumothoraces or unilateral pleural effusions despite negative imaging and negative pleural fluid studies. Pleural biopsy may be necessary early in the workup in recurrent hydro pneumothoraces or unilateral pleural effusions in elderly with a history of asbestos exposure.

Conflicts of interest

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