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Case report

Malignant pleural mesothelioma in a 17-year old boy: A case report and literature review

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ABSTRACT

Background: Malignant pleural mesothelioma is a rare, invasive and often fatal neoplasm that develops in the thin layer of tissue surrounding the lungs known as the pleura. Although rare, mesotheliomas do occur in the young; their characteristics are distinct from those of older patients.
Case presentation: This is a case report of a 17-year-old boy who had moderate dyspnea, cough, right-sided pleuritic chest pain, fever, headache and no weight loss. Physical examination showed a right pleural effusion and chest roentgenograms revealed a homogenous opacity on lower right hemithorax. Biochemical analysis of pleural fluid showed hemorrhagic/turbid effusion compatible with exudate. It was initially treated as an empyema. The pleural fluid culture was negative. Adenosine deaminase level was 34.3 U/L (admission) and 19.02 U/L (two weeks after). Pleural fluid smear and culture for *Mtb* were negative. During the open pleural biopsy, thickened pleura and multiple pale yellow nodules in the lung were observed. The histopathological report was compatible with malignant pleural mesothelioma. With this diagnosis, a chemotherapy regimen with cisplatin was initiated. After two cycles, the patient had no clinical and radiological improvement. The patient is currently under regular follow up.
Conclusion: MPM is rare in young adults and its clinical presentation makes it different from mesothelioma in elderly patients, so it will be necessary to identify the new risk factors that can identify these patients.

1. Background

Malignant pleural mesothelioma (MPM) is a rare, aggressive and hard-to-treat malignant neoplasm that commonly develops in pleura and peritoneum [1]. Approximately 80% of MPM patients have a history of exposure to asbestos [2]. However, it can take decades to develop and, as a result, is usually thought of as a disease of middle age and elder people [2]. Patients with MPM frequently develop thoracic pain, dyspnea, weight loss and pleural effusion [3]. Unusual presentations have also been reported, though rarely [4,5]. Although rare, pleural mesothelioma does occur in the young; their characteristics are different from those of older patients [6]. We report a case of a teenager who had a large malignant pleural effusion.

2. Case presentation

A 17-year old male patient was admitted to our hospital on June 2015. He had smoked tobacco and marijuana cigarettes for three months the previous year, as well as moderate alcohol consumption. The patient was diagnosed with allergic rhinitis and bronchial hyperreactivity at the age of 12 years old without regular treatment. At hospital admission, he had 7 days with fixed moderate dyspnea, fever, right-sided pleuritic chest pain, headache, cough and no weight loss. Physical examination revealed a pleural effusion syndrome on the same side. No other associated clinical finding was described. Chest x-ray showed a homogenous opacity in lower right hemithorax (Fig. 1). A chest tube was inserted and 1600 ml of hemorrhagic/turbid pleural fluid was drained. After we diagnosed complicated parapneumonic pleural effusion, Clindamycin and ceftriaxone were given. Several biochemical and microbiological analyzes were performed.

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The next day, the patient had relief of symptoms, reporting mild pain only at the chest-tube insertion site. A second chest X-ray showed a fully re-expanded left lung (Fig. 2). For the next 7 day at least 750 ml daily of clear pleural fluid was drained. Biochemical analysis of pleural fluid demonstrated an exudate with very low glucose level 0.20 mg/dl. Pleural fluid cultures were negative. Because of the increase of pleural fluid drainage up to 800 ml daily, we decided to perform a closed pleural biopsy, which reported negative results for tuberculosis and malignancy.

At admission, the adenosine deaminase (ADA) level was 34.3 U/L in two weeks it decreased to 19.02 U/L. Acid-fast bacilli (AFB) for pleural fluid was negative, and culture for *Mtb* remained negative.

In spite of antibiotics, pleural fluid drainage did not decrease after 3 weeks, thus we decided to begin the anti-TB treatment. However, after another week, no decrease in pleural fluid drainage was observed. Therefore, we decided to perform an open pleural biopsy. During surgery, pleural thickening and multiple pale yellow nodules in the lung were seen. After a diagnosis of malignancy, pleurodesis was done twice with povidone-iodine but no decrease in pleural fluid drainage was obtained.

Unfortunately, the patient did not receive other treatment such as pemetrexed, gemcitabine because they were not available at that moment, and extrapleurectomy is not a common surgical procedure in our media.

Immunohistochemistry revealed tumor cells to be immunoreactive for calretinin (*clone Z11-E3, Dako*) cytokeratin 7 (*BC-1, BIOCARE MEDICAL*) and negative for WT1 (*BC.6F–H2, BIOCARE MEDICAL*) NAPSIN A (*TMU-Ad 02, BIOCARE MEDICAL*). Based on these findings; the cause of pleural effusion was diagnosed as MPM. With this diagnosis, a chemotherapy regimen with cisplatin was initiated [7]. After two cycles, there was no clinical and radiological improvement. The patient is currently under regular follow up.

3. Discussion

The worldwide MPM incidence has been rising since the mid 20th century [8,9]. MPM is often considered a disease of the elderly with a median age of 74 years [10]. However, there is a subgroup of patients who are young [6]. Here, we showed the case of a teenager with massive pleural effusion, diagnosed with MPM and a review of the literature. As far as we know, this is one of the few younger patients with MPM reported until now.

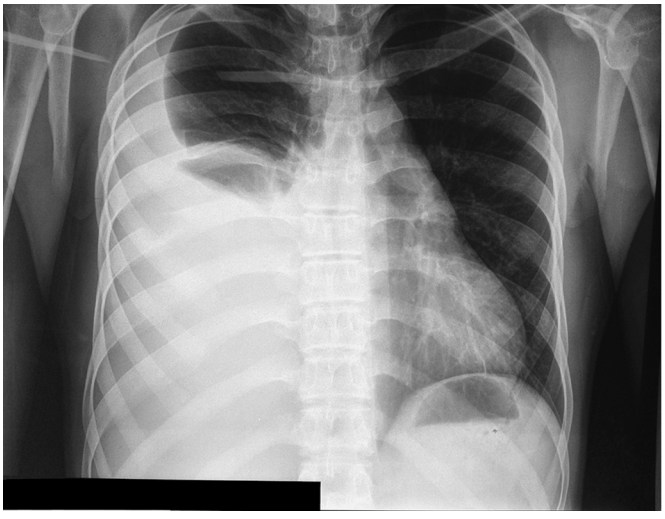

Fig. 1. Initial Chest X-ray demonstrated a large right pleural effusion, that occupies 2/3 parties of the right hemithorax accompanied with fever and right chest pain.

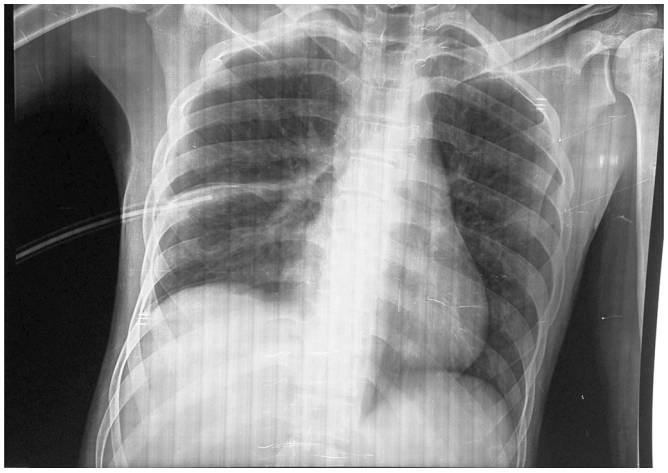

Fig. 2. Chest X-ray demonstrated total lung reexpansion after thoracic tube insertion. Mild pleural thickness is observed.

To date, numerous epidemiological reports describe the elderly population with MPM but outside of case studies [11,12], and small cohorts [13,14] a detailed examination of young patients with MPM has been less studied. An important finding of these reports is that almost 15**%** of cases with MPM were younger than the age 40 [14–17]. In addition, other case-series have also found a lower frequency for patients with MPM under 40 years old: i.e. Kane et al. found ten of 172 cases (5.8%) [18], whereas Jones and Thomas in a review of 5778 death certificates reported 298 cases (5.2%) between the ages of 15 and 44 at the time of death [19]. The Surveillance, Epidemiology, and End Results (*SEER*) database for mesothelioma cases from 1990 to 2010 showed that about 2% (207) of the 12,345 mesothelioma cases studied occurred in people under 40 years old [20].

Malignant mesothelioma (MM) in young people acts very differently than it does in the older population. Thus, while almost 80% of the older MPM patients are men, the disease is about equally split between men and women in the younger age group [21]. Another difference between younger and older mesothelioma patients is the frequency of pleural and peritoneal mesothelioma. The two types of mesothelioma are about evenly distributed in the younger patients while 90% of older patients have the pleural variety [21]. The younger group is also more likely to undergo cancer-directly to surgery and to live longer afterward, even if they have a type of mesothelioma that is usually considered less survivable [22,23].

Most of the elderly patients diagnosed with mesothelioma die of the disease within a year of diagnosis. Improved overall survival of the young mesothelioma patients with both pleural and peritoneal mesothelioma indicates that mesothelioma in the young may possibly have a different natural history and indeed be biologically different from mesothelioma in elderly patients. Genomic and expression studies of tumor samples from young and old patients with mesothelioma will clarify the biological differences between the two cohorts. To our knowledge, no such studies have been conducted to date.

The relationship between MM and asbestos exposure it is well-known. The latency period of MM is highly variable and can be as short as 13 years or as long as 70, between exposure and diagnosis [24]. Because of this long time span, most of those who are diagnosed with MPM and other asbestos-related disease are in their 60s or 70s.

MPM in young patients with occupational and non-occupational