Massive malignant pleural effusion due to lung adenocarcinoma in 13-year-old boy

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Abstract
A 13-year-old boy with no risk factors for lung cancer presented with a massive left-sided pleural effusion and a mediastinal shift on chest radiography and computed tomography. A chest tube drained bloody pleural fluid with an exudative pattern. A pleural biopsy and wedge biopsy of the left lower lobe revealed mucinous adenocarcinoma in the left lower lobe wedge biopsy and metastatic adenocarcinoma in the pleural biopsy. The patient is currently undergoing chemotherapy. Radiotherapy is planned after shrinkage of the tumor. Adenocarcinoma of the lung is very rarely seen in teenagers or children, especially in the absence of risk factors.

Keywords
Malignant, Pleural effusion, Lung, Adenocarcinoma

Introduction
Adenocarcinoma of the lung arises from atypical adenomatous hyperplasia progressing to bronchioloalveolar carcinoma which then transforms into invasive adenocarcinoma. This tumor is the most common type of lung cancer in women and nonsmokers, but the average age of patients is 45 years, and it is very rarely seen in teenagers or children.¹,² The prognosis of primary adenocarcinoma in adults and children is equally poor.³ There is a strong relationship between risk factors (smoking, underlying infections, air pollution, genetic) and adenocarcinoma, but in rare cases, none of these risk factors are detected.

Case report
A 13-year-old boy, who was treated for cough, coryza, chills and fever for one month with a poor response to medical therapy, presented with a massive left-sided pleural effusion and a mediastinal shift on chest radiography and computed tomography (Figure 1). He had no history of trauma or family history of cancer, and he was not an active or passive smoker. He was tachypneic (28 breaths min⁻¹) but afebrile. On physical examination, respiratory sounds were absent on the left side. A chest tube as inserted and he was transferred to the intensive care unit. Because of unilateral bloody pleural fluid with an exudative pattern (Table 1), thoracoscopy was performed via a camera inserted into the chest tube hole. We observed a hepatized lung with severe pleural inflammation, thus a lateral minithoracotomy was performed for pleural biopsy and wedge biopsy of the left lower lobe (Figure 2). Pathology revealed mucinous adenocarcinoma in the left lower lobe wedge biopsy and metastatic adenocarcinoma in the pleural biopsy (Figure 3). Because of the family’s low economic status, we could not perform immunohistochemistry. Smear and culture of the pleural fluid were negative for tuberculosis. Cytology of the pleural fluid was...
positive for malignancy. Some tumor markers were elevated (Table 2). Fiberoptic bronchoscopy after expansion of the lung showed no endobronchial lesion. Metastatic evaluations, including brain and abdominopelvic computed tomography and a bone scan, were negative. Chemotherapy with cisplatin and gemcitabine was started. The patient has completed the 4th course and 2 additional courses remain. Radiotherapy is our next plan after shrinkage of the tumor. So far, his condition is good and the response to treatment is acceptable.

Discussion

Primary pulmonary neoplasms are extremely rare in children, and metastatic tumors of nonpulmonary origin (osteogenic sarcoma, Wilms tumor, and rhabdomyosarcoma) are more prevalent than primary lung cancers. Benign tumors constitute the majority of primary pulmonary neoplasms. The clinical presentation of lung cancer includes local symptoms that can be bronchopulmonary, non-bronchopulmonary, or from metastatic disease, and may also be due to paraneoplastic syndrome. Symptoms can mimic a common cold or pneumonia, and chest radiographs may be similar to pneumonia consolidation of the lung. In every patient who receives medication for pneumonia, the diagnosis should be established by chest radiography. Recurrent pneumonia and failure to improve with medical treatment should raise suspicion of underlying problems such as malignancy. The prognosis for primary malignancy in children is best for endocrine or mucoepidermoid tumors, but as in adults, the outcome for adenocarcinoma is poor, especially in patients with metastatic disease where surgery is excluded from multimodality therapy. Primary lung adenocarcinoma in children frequently presents as metastatic disease at diagnosis, and the median survival is 6–9 months.

Immunohistochemistry can help to confirm the lung as the primary source of metastatic adenocarcinoma. In our patient, because of the family's economic status, we could not perform positron-emission tomography for...
staging, immunohistochemistry for definite diagnosis, or molecular testing to establish the histologic subtype and decide systemic therapy. We started 6 courses of gemcitabine (1000 mg m$^{-2}$C0$_{2}$C1 day on days 1 and 8) plus cisplatin (75 mg m$^{-2}$C0$_{2}$C1 day on day 1) with a week between courses. At the end of 4th course, the tumor

Figure 2. (a) Chest radiograph, (b) parenchymal view of the left upper lobe, (c) mediastinal view of the left lower lobe, and (d) parenchymal view of the left lower lobe after thoracotomy.

Table 2. Serum tumor markers.

<table>
<thead>
<tr>
<th>Tumor marker</th>
<th>Value</th>
<th>Normal range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carcinoembryonic antigen</td>
<td>13.1</td>
<td>3.5–5</td>
</tr>
<tr>
<td>Carbohydrate antigen 19-9</td>
<td>318.4</td>
<td>0–35</td>
</tr>
<tr>
<td>Beta-human chorionic gonadotropin</td>
<td>1.5</td>
<td>0–15</td>
</tr>
<tr>
<td>Alpha fetoprotein</td>
<td>1</td>
<td>0.2–8.8</td>
</tr>
<tr>
<td>Beta-2 microglobulin</td>
<td>1.1</td>
<td>≤ 3</td>
</tr>
</tbody>
</table>

Figure 3. Pathology revealing a mucinous adenocarcinoma: hyperchromic nuclei with mitosis and atypia. Hematoxylin and eosin stain, original magnification ×40.
response to chemotherapy was acceptable. More follow-up after completion of treatment is needed to document final survival.

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