SHORT COMMUNICATION

Apparent primary pleural melanoma: case report and literature review

S. W. Um, C. G. Yoo, C. T. Lee, S. K. Han, Y. S. Shim, and Y. W. Kim

Department of Internal Medicine and Lung Institute of Medical Research Center, Seoul National University College of Medicine, and Clinical Research Institute, Seoul National University Hospital, 28 Yongon-Dong, Chongno-Gu, Seoul 110-774, South Korea

INTRODUCTION

Melanoma has been noted to occur in most systemic tissues, both as a primary and a metastatic lesion. In the English literature, only one case of primary pleural melanoma has been reported by Smith and Opipari in 1978. (1) The case history is presented of a patient who developed apparent primary pleural melanoma and consequently severe dyspnea.

CASE REPORT

A 61-year-old male presented with dyspnea. He had been well, until diagnosed at another institution with pulmonary tuberculosis, 12 months previously. Following this diagnosis, he was treated with antituberculosis chemotherapy for 9 months. One month before admission to our hospital, dyspnea and chest pain developed. He was initially admitted to another facility and chest radiographs revealed a left-sided pleural effusion. The patient repeatedly required therapeutic pleural fluid drainages and chest tube insertion without clinical improvement due to the rapid accumulation of fluid. A flexible, fiberoptic bronchoscopy performed at another institution revealed no abnormal findings. Following referral to our hospital for further evaluation, the patient on examination had a blood pressure of 120/70 mmHg, a pulse of 100/min, a temperature of 37°C and respiration rate of 20/min. Chest examination revealed diminished breath sound and dullness to percussion in the left lung. He denied having had spontaneous regression of skin lesions or treatment of moles by excision or cautery. He had lost 8 kg in the previous 1 year. The skin of the entire body and scalp was carefully inspected for possible sites of melanoma with no result except for a senile lentigo on the face, which was confirmed by a dermatologist. Results of rectal and genital examinations were normal.

A chest radiograph at admission demonstrated a huge lobulating mass in the left hemithorax. A contrast enhanced computed tomographic scan of the chest (Fig.1) displayed the pleura-based mass containing fluid and involving the diaphragm in the left hemithorax, but no evidence of lesion in the right hemithorax. An endoscopic examination showed no abnormalities in the esophagus. Visualization of fundi disclosed no abnormalities. Repeated cytologic examinations of pleural fluid demonstrated malignant cells with black pigments. On the 7th hospital day, he underwent a video-assisted thoracoscopic biopsy. Thoracoscopy revealed a huge black-pigmented pleura-based mass. Immunohistochemical stainings of the specimens were strongly positive for S-100 and HMB-45; findings that are characteristic of malignant melanoma.

Dyspnea and weakness progressed rapidly to the point where he could not expectorate efficiently and had to receive a tracheostomy for the removal of the sputum on the 26th hospital day. The patient and family refused chemotherapy and on the 29th hospital day, the patient transferred himself to another hospital for supportive management.

DISCUSSION

Since melanoma metastasizes frequently to the lung and much less frequently to the pleura, (2), any intrathoracic lesion should be assumed to be metastatic until proved otherwise.

Jensen and Egedorf (3) proposed the following six clinical criteria for the diagnosis of primary pulmonary melanoma: (1) no previously removed pigmented skin tumors, (2) no ocular tumors removed, (3) a solitary tumor in the surgical specimen, (4) tumor morphology compatible with a primary tumor, (5) no demonstrable melanoma in other organs at the time of operation, and

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Correspondence should be addressed to Young Whan Kim, MD, PhD
Fax: +82-2-762-9662 E-mail : ywkim@snu.ac.kr
(6) autopsy without primary malignant melanomas being demonstrated elsewhere. These criteria should also hold well for primary pleural melanoma.

However, the manner in which melanoma can develop as a primary neoplasm in the pleura remains unclear. Some investigators have suggested that extracutaneous melanoma may arise from residual melanoblasts (3–8). It has been demonstrated that melanoblasts arise from the neural crest and migrate to all parts of the body. Melanoblasts have been identified in the normal vagina and esophagus (7,8), which supports the suggestion that melanoma can arise in these areas. If melanomas occur primarily in the pleura, melanoblasts should also be demonstrable in the pleura. But, no such study has been performed on the pleura.

Other investigators have hypothesized that extracutaneous melanomas arise in areas of squamous metaplasia (4,5,9). In the case presented by Salm (4), stratified squamous epithelium, identical in appearance to cutaneous melanoma, was adjoining the main tumor. However, the specimens of our patient did not manifest squamous metaplasia.

Since melanoma of the skin often regresses spontaneously (10), it is possible that this “primary” pleural melanoma may be a metastasis from an occult primary lesion. However, our patient had no history of suspicious skin lesions or moles and the inspection of the skin of whole body revealed no abnormalities except for a senile lentigo on the face. Nonetheless, this case did not meet the clinical criteria proposed by Jensen and Egedorf, because the patient was still alive at discharge and an autopsy was not performed. However, surgical specimens of our patient were typical of malignant melanoma and clinical, radiologic, bronchoscopic and thoracoscopic findings indicated a huge pleura-based mass confined to the left hemithorax without involvement of the airways, which suggested that the lesion was a primary pleural lesion rather than a metastatic one.

In conclusion, this case can be designated apparent primary pleural melanoma, due to limitations beyond our control, i.e. lack of autopsy findings and no information on the presence of melanoblasts in the pleura. In the future, further study is required for the presence of residual melanoblasts in the pleura.

REFERENCES