

An Autopsy Case Report of Malignant Pleural Mesothelioma with Deciduoid Features

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Abstract

Deciduoid mesothelioma is a rare variant of epithelioid mesothelioma. We experienced the case of a 73-year-old man with asbestos exposure who was diagnosed with malignant pleural mesothelioma with deciduoid features. He received chemotherapy containing six cycles of cisplatin and pemetrexed and survived for twenty-five months after the diagnosis. At autopsy, the final diagnosis was biphasic pleural mesothelioma. Cells with deciduoid features had mostly disappeared, and spindle cells markedly proliferated. To the best of our knowledge, this is the first autopsy case of malignant pleural mesothelioma with deciduoid features that exhibited a response to chemotherapy.

Key words: mesothelioma, deciduoid, pleura, autopsy

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Case Report

A 73-year-old man visited our hospital with sudden onset dyspnea and left chest pain. He quit a 40 pack-years smoking habit at 63 years of age when he received pancreatoduodenectomy for an adenocarcinoma of the major duodenal papilla. He also had a history of asbestos exposure when he worked as a shipyard worker until retirement. A chest X-ray examination revealed a collapse of the left lung. Subsequent computed tomography of the chest revealed the collapse of the left lung, diffuse distribution of a low attenuation area in both lungs and trivial thickening at the dorsal region of the left parietal pleura, leading to a diagnosis of secondary spontaneous pneumothorax. During partial lobectomy under video-assisted thoracic surgery for the continuous air leakage with catheter drainage, biopsy of a white parietal pleura lesion was also performed. A histological analysis of the specimens revealed that polygonal or round-shaped cells with abundant eosinophilic and glassy cytoplasm prolifer-

ated like a sheet. Some of these cells possessed round, vesicular nuclei (Fig. 1A, B). An immunohistochemical examination of the tissue showed positivity for calretinin (Fig. 1C), D2-40 (podoplanin), Wilms' tumor-1 (WT-1), and cytokeratin 5/6 (CK 5/6) but negativity for thyroid transcription factor-1 (TTF-1). Elastica van Gieson staining and hematoxylin and eosin staining of the lung showed rupture of the visceral pleura with a deficit of elastic fibers beside the tumor cells, which indicated the tumor induced pneumothorax. From these abovementioned features, the diagnosis of secondary pneumothorax caused by a malignant pleural mesothelioma with deciduoid features was confirmed.

The patient received first-line chemotherapy with six cycles of cisplatin (CDDP, 75 mg/m², day 1) and pemetrexed (PEM, 500 mg/m², day 1). After chemotherapy, the lesions showed a partial response and decreased in size from 11.7 mm to 7.9 mm (Fig. 2A, B). Subsequently, the patient received four cycles of PEM (500 mg/m², day 1) alone. After chemotherapy with PEM, the tumor enlarged from 7.9 to 9.25 mm (Fig. 2B, C). The patient subsequently received

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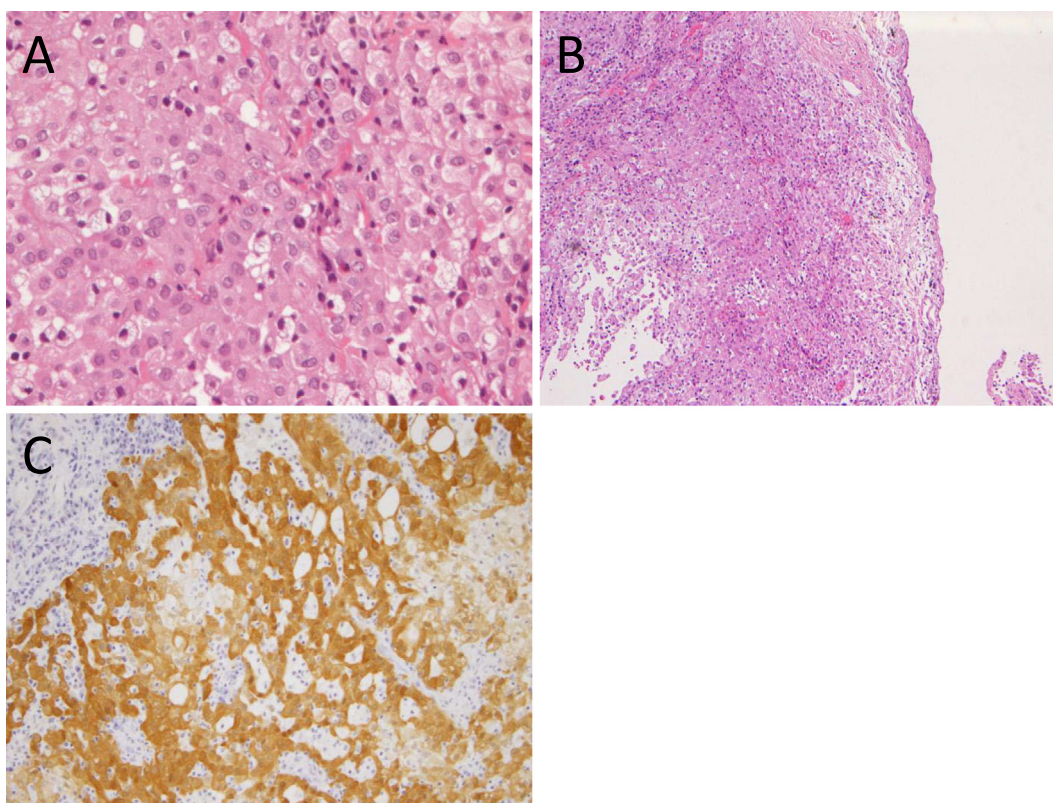


Figure 1. Photomicrographs of partial lobectomy. Cells have deciduoid features, large, polygonal, round, or ovoid cells with well-defined cell borders, abundant eosinophilic glassy cytoplasm, and round vesicular nuclei. Some cells present prominent nucleoli [Hematoxylin and Eosin (H&E) staining; A: 400 $\times$, B: 100 $\times$]. Immunohistochemical staining showed calretinin (C: 200 $\times$) positivity.

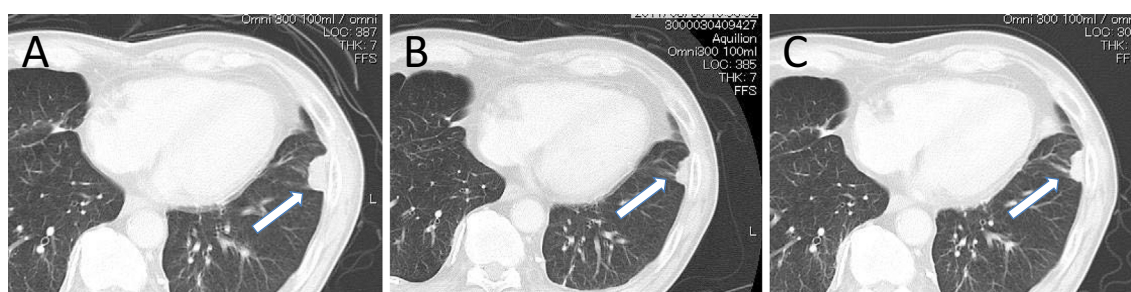


Figure 2. Computed tomography at the start of chemotherapy (A), after first-line chemotherapy consisting of CDDP and PEM (B) and after second-line chemotherapy with PEM alone (C). The pleural tumor thickness decreased from 11.7 to 7.9 mm after first-line chemotherapy (A, B). After second-line chemotherapy, the tumor thickness enlarged from 7.9 to 9.25 mm (B, C).

five cycles of platinum, one course of CDDP (75 mg/m², day 1) and four courses of carboplatin (CBDCA, area under the concentration curve =5, day 1), and PEM (500 mg/m², day 1). Substitution of CDDP to CBDCA was due to mild renal dysfunction after CDDP readministration. Thereafter, the progression of the lesions was observed, and two cycles of gemcitabine alone (GEM, 1,000 mg/m², days 1 and 8) was administered without tumor regression. Three months after the last chemotherapy treatment, the patient died due to tumor progression. The progression-free survival of the first-line chemotherapy consisting of CDDP and PEM, and subsequent chemotherapy with PEM alone, was approximately

ten months. In total, the patient received chemotherapy for twenty-two months and survived for twenty-five months after the diagnosis.

An autopsy was performed sixteen hours after his death. The final diagnosis from the specimens obtained at autopsy (Fig. 3) was biphasic pleural mesothelioma. Deciduoid features were only visible in a limited area of the primary lesion, and spindle cells markedly proliferated. Approximately half of the tumor cells with deciduoid features necrotized. A histological evaluation of the efficacy of chemotherapy was classified as Ef 1b (1). The tumor directly invaded the left lung and mediastinum. Metastasis of the disease was ob-

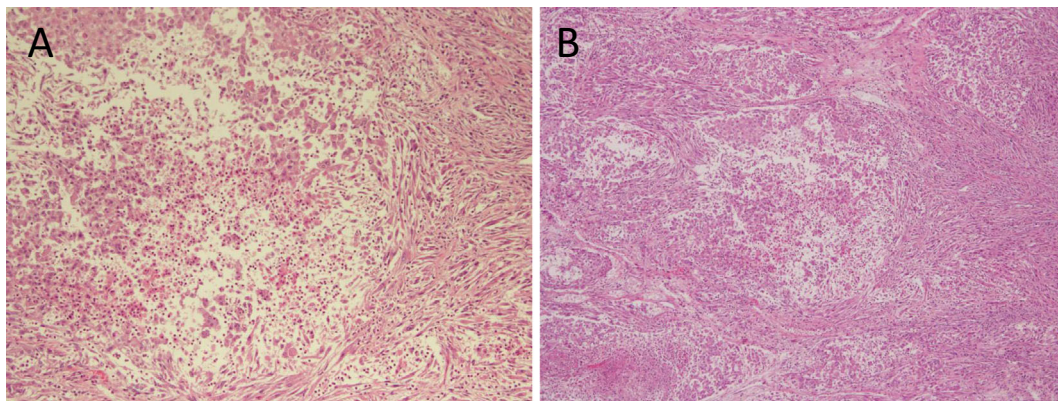


Figure 3. Photomicrographs at autopsy. Deciduoid features were only seen in a limited area of the primary lesion and spindle cells had markedly proliferated, which shows biphasic mesothelioma. Approximately half of cells that had deciduoid features had necrotized (H&E staining, A: 100×, B: 50×).

served in the right lung, liver, left kidney, vertebra, and epicardium. Pulmonary emphysema and anthracosis were also visible due to smoking.

Discussion

Deciduoid mesothelioma is a relatively rare variant of epithelioid mesothelioma that is characterized by the presence of deciduoid morphological features, such as very large, round or polygonal epithelioid cells with abundant eosinophilic, glassy cytoplasm arranged in solid nests or trabecula (2). Deciduoid mesothelioma was first reported in the peritoneum of a young woman without asbestos exposure (3). The first description of the subtype of pleural mesothelioma was reported in 2000 (4). Only forty cases of deciduoid pleural mesothelioma have been reported (2, 5-7). Previously, it was thought to be limited to the peritoneum and women with no known history of asbestos exposure. However, several cases of the tumor in the pleural cavity of aged men with asbestos exposure have been recently described (2). As with previous reports, our case of this type of mesothelioma was aged and had been exposed to asbestos.

Chemotherapy comprising CDDP and PEM is the standard of care for malignant pleural mesothelioma. However, only two cases of malignant deciduoid pleural mesothelioma treated with CDDP and PEM chemotherapy have been reported. In one case, the tumor showed progression after three cycles of chemotherapy and the patient died three months after the initial diagnosis (5). In another case, chemotherapy with CDDP and PEM yielded a good response after five cycles. Subsequent radical surgery and another three cycles of chemotherapy was performed, and the patient ultimately survived for two years (6). In the present case, the patient received chemotherapy with six cycles of CDDP and PEM, and the lesion temporarily showed slight regression. The patient ultimately survived for twenty-five months

after the diagnosis. In the specimens at autopsy taken after courses of chemotherapy, cells with deciduoid features had nearly disappeared and approximately half of the cells showed necrosis, in contrast to the marked proliferation of spindle cells. These findings indicate that deciduoid mesothelioma is able to respond to chemotherapy comprising CDDP and PEM, similar to the epithelioid type but not the sarcomatoid type.

To the best of our knowledge, this is the first autopsy case of malignant pleural mesothelioma with deciduoid features and the potential efficacy of chemotherapy was confirmed. However, the efficacy of chemotherapy for this rare subtype of tumor must be assessed in additional cases.

The authors state that they have no Conflict of Interest (COI).

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