

Sclerosing pneumocytoma with metastasis to the mediastinal and regional lymph nodes

Xiaojun Wang, Lizhi Zhang¹, Yanfu Wang, Xuzhao Jia, Jiali Wang, Hua Zhang

Departments of Geratology and ¹Pathology, First Affiliated Hospital, Dalian Medical University, Dalian, China

Address for correspondence:

Prof. Hua Zhang, Department of Geratology, First Affiliated Hospital, Dalian Medical University, 222 Zhongshan Road, Xigang District, Dalian 116011, Liaoning Province, China. E-mail: zhanghua2233@126.com

ABSTRACT

Sclerosing pneumocytoma (SP) is an uncommon benign tumor, and metastasis of SP has been rarely reported. Here, we report the case of a 26-year-old woman with surgically confirmed SP. The tumor diameter was 40 mm, and metastasis to mediastinal and regional lymph nodes was observed. Immunohistochemically, both surface and round cells were positive for epithelial membrane antigen, thyroid transcription factor 1, and vimentin. Only surface cells expressed creatine kinase, carcinoembryonic antigen, napsin A, and cytokeratin 7, and only round cells expressed progesterone receptor. Ki-67 was detected in ~3% of cells, and the rate of weak positive p53 staining was 3%. Both cell types were negative for chromogranin A, synaptophysin, CD3, and CK20. Multiple metastases in a young SP patient are very rare, and potential mechanisms of metastasis may be related to epithelial-mesenchymal transformation.

KEY WORDS: Immunohistochemistry, lymph node metastasis, sclerosing pneumocytoma, vimentin

INTRODUCTION

Sclerosing pneumocytoma (SP) is an uncommon benign tumor, and some cases of SP metastasis have been reported. However, reports of SP with multiple lymph node (LN) metastases are very rare. Here, we report the case of a young female patient with SP and interlobar, hilar, and mediastinal LN metastasis and provide an analysis of the immunohistochemical characteristics to explore the mechanisms of SP metastasis.

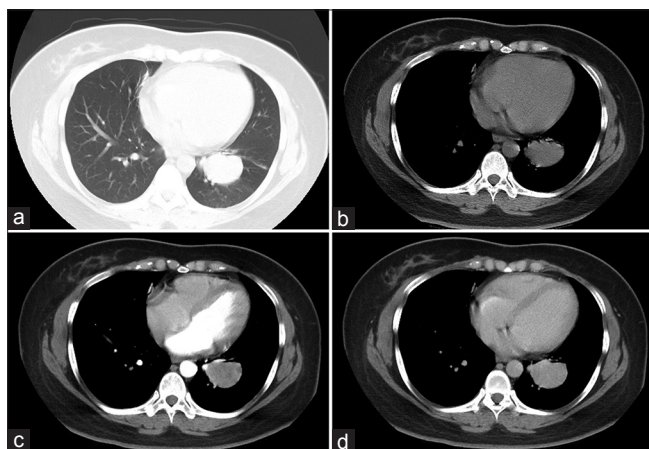


Figure 1: Computed tomography findings. (a) A mass was observed in the anterior basal segment in the left lower pulmonary field, with a clear boundary and irregular margins. (b) No swelling of mediastinal or hilar lymph nodes and no bilateral pulmonary effusion were observed. (c, d) Enhanced CT showed uniform enhancement in the venous phase and arterial phase

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CASE REPORT

A nodule was found in a 26-year-old woman on a chest X-ray during an employment health examination. She had a 10-year smoking history but no cough, hemoptysis, chest pain, or other pulmonary symptoms. No increases in the relevant tumor markers were observed.

Chest computed tomography scanning revealed a mass in the anterior basal segment in the left lower pulmonary field with a clear boundary and irregular margins. No swelling of the mediastinal or hilar LNs was observed [Figure 1]. The patient underwent pulmonary lobectomy of the left lower lobe with the removal of Group VII, X, and

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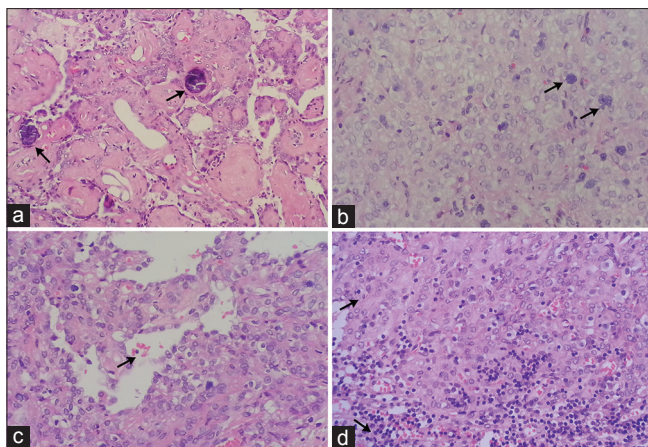


Figure 2: Hematoxylin and eosin staining showed (a) calcification in an interstitial sclerosis pattern (black arrows, $\times 200$). (b) The cells in solid patterns were round with infiltration of mast cells (black arrows, $\times 400$). (c) Some red blood cells had irregular lacunar in hemorrhagic patterns (black arrows, $\times 400$). (d) The architecture of the lymph node was partially missing and replaced with mainly round cells (black arrows, $\times 400$)

XI LNs. Pathological examination revealed metastasis in 1 of 3 Group VII nodes, 1 of 1 Group X node, and 0 of 2 Group XI nodes as well as in 3 of 4 interlobar nodes. She received no chemotherapy and remained healthy during 9 months of follow-up.

On pathological examination, grossly, the tumor was 40 mm \times 40 mm \times 30 mm in size and sharply demarcated from the surrounding lung tissue. The cut surface was gray and whitish. Microscopically, the tumor consisted of both surface and round cells. The tumor included solid, papillary, sclerosing, and hemorrhagic patterns. Some papillary interstitial sclerosis and calcification were found. The cells in solid patterns were round, with infiltration of mast cells. The architecture of the LNs was partially missing and had been replaced by abnormal proliferation of cells, mainly including stromal round cells and only a few cuboidal surface cells [Figure 2].

Immunohistochemically [Figure 3], both the surface and round cells were positive for epithelial membrane antigen, thyroid transcription factor 1, and vimentin. The surface cells stained positively for creatine kinase, carcinoembryonic antigen, napsin A, and cytokeratin 7. The round cells stained positively for progesterone receptor. Ki-67 was detected in approximately 3% of the cells, and the rate of weak positive p53 staining was 3%. Both cell types were negative for chromogranin A (CgA), synaptophysin (Syn), CD3, and CK20. The pathologic diagnosis was SP with no tumor involvement in the bronchial stump and some LN metastasis in the mediastinal, hilar, and mediastinal LNs (1/3, 1/1, and 3/4, respectively).

Publication of this case was approved by the Ethics Committee of the First Affiliated Hospital of Dalian Medical University, and the patient provided written informed consent for publication of the case and accompanying images.

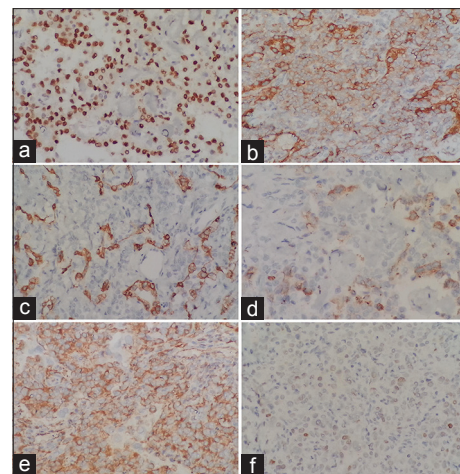


Figure 3: Immunophenotyping findings. (a) Both surface and round cells stained positively for thyroid transcription factor 1. (b) Surface cells showed strong expression of epithelial membrane antigen, and round cells showed weak expression of epithelial membrane antigen. The surface cells stained positively for (c) creatine kinase and (d) napsin A. (e) Both the surface and round cells stained positively for vimentin. (f) Weak positive staining for p53 was observed in approximately 3% of cells ($\times 400$)

DISCUSSION

Based on the previously reported cases, SP transfer may be related to age, gender, primary tumor location, and tumor size, and metastasis may be more likely to occur in young patients, males, and those with large tumors and tumors in the lower left lung.^[1,2] The mechanism of SP transfer remains unclear.

SP tumors are composed of two slightly different histogenetic cell types, cuboidal surface cells, and stromal round cells. Both are considered to be neoplastic and derived from primitive respiratory epithelial cells.^[3] However, round cells are not as mature as cuboidal cells.^[4] The surface cells resemble Type II pneumocytes. The round cells have been found to express neuroendocrine markers, i.e., CgA, neuron-specific enolase, and Syn and mesenchymal marker vimentin.^[5] Metastatic deposits in the LN were found to consist of round cells,^[1] and in our case, we also found that mainly round cells are in the metastatic LN. Sun *et al.*^[6] argued that round cells may be derived from epithelial-mesenchymal transformation (EMT) of the surface cells. EMT could be closely involved in metastasis of cancer.^[7] In the present case, expression of epithelial markers was observed only in surface cells, and the mesenchymal marker vimentin expression was found in both surface and round cells. Vimentin plays important roles in epithelial cell migration and invasion,^[8] and the multiple metastases in our patient were likely related to high levels of vimentin expression. Although vimentin expression was investigated in only four previous cases of SP transfer, in all four cases, round cells were positive for vimentin expression.^[2,9-11] In our analysis, the potential SP transfer mechanisms may be related to EMT.

Notably, even though our patient had multiple metastases, the prognosis was not influenced, and surgery was an effective treatment.

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Conflicts of interest

There are no conflicts of interest.

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