Yonsei Medical Journal
Vol. 47, No. 4, pp. 571 - 574, 2006

Lung Metastasis from an Immature Teratoma of the Nasal Cavity Masquerading as Small Cell Carcinoma of the Lung

Min Kyu Kang,[1] Yong Chan Ahn,[1] Joon Oh Park,[2] Joungho Han,[3] and Kyung Soo Lee[4]

Departments of [1]Radiation Oncology, [2]Medicine, [3]Pathology, and [4]Radiology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, Korea.

We report a case of small cell lung cancer that turned out to be a metastatic teratoma from the nasal cavity rather than a new primary cancer. A 54-year-old woman was diagnosed with an immature teratoma of the nasal cavity with a predominant neuroblastomatous component. Small cell lung cancer was detected by bronchoscopic biopsy 21 months later, and it was treated with concurrent radiochemotherapy as if it had been a new primary cancer. Since a recurrent tumor containing fat- like density grew slowly on the serial chest CT scans after achieving complete response, we reached the conclusion that the small undifferentiated cells could be metastatic neuroblastomatous components from the immature teratoma of the nasal cavity.

Key Words: Teratoma, small cell carcinoma, metastasis

INTRODUCTION

In 1984, Heffner first described a uncommon tumor of sinonasal teratocarcinosarcoma (SNTCS), which has the variegated microscopic features of non-germ cell malignant components such as adenocarcinoma, rhabdomyosarcoma or neuroblastoma.[1] Non-germ cell malignant components arising from SNTCS have metastatic potential, and if a metastatic tumor from non-germ cell malignant components is detected before the primary SNTCS is detected, then it could be misdiagnosed as a primary or metastatic tumor of a non-germ cell tumor.[2,3] Herein, we report a case that presented as a small cell carcinoma of the lung that was suspected of being a metastatic tumor rather than a new primary cancer in a patient who had a past history of SNTCS of the nasal cavity.

Received January 31, 2005
Accepted April 28, 2005

Reprint address: requests to Dr. Yong Chan Ahn, Department of Radiation Oncology, Samsung Medical Center, Sungkyunkwan University School of Medicine, 50 Ilwon-dong, Gangnam-gu, Seoul 135-710, Korea. Tel: 82-2-3410-2602, Fax: 82-2-3410-2619, E-mail: ycahn@smc.samsung.co.kr

CASE REPORT

In February 1998, a 54-year-old Korean woman visited a local clinic presenting with nasal obstruction. A mass in the right nasal cavity was resected under local anesthesia without suspicion of malignancy and histopathologic diagnosis of immature teratoma with a predominant neuroblastomatous component was made. Magnetic resonance imaging after resection showed neither remnant nor metastatic lesions. In May 1998, a gray- to black-colored mass with a foul odor regrew in her right nasal cavity. The recurrent tumor was excised and again showed immature teratoma with a neuroblastomatous component (Fig. 1). Adjuvant radiation therapy to the nasal cavity was administered (60 Gy over 6 weeks, at a daily dose of 2 Gy) and there was no evidence of recurrence until May 1999.

In October 1999, she presented with a cough, and subsequent chest computed topography (CT) scans showed two pulmonary masses in the right upper lobe with mediastinal lymphadenopathy. Bronchoscopy revealed a beefy mass obstructing the right upper lobar bronchus, and the biopsy specimen showed malignant tumor cells characterized as compact sheets of small round cells, hyperchromatic nuclei, inconspicuous nucleoli associated with squeezing artifacts, and a scant amount of cytoplasm; there were no other mature

epithelial components or adipose tissue (Fig. 2). All the systemic staging work-ups were negative. These findings led us to the conclusion that the lung mass was a new primary small cell carcinoma of the lung. She was treated with concurrent radiochemotherapy (CRCT) from December 1999 to March 2000, which consisted of thoracic radiation therapy (TRT) of 44 Gy over 4.5 weeks (at a daily dose of 2 Gy) and four cycles of chemotherapy with intravenous cisplatin and oral etoposide; the first two cycles were delivered concurrently with the TRT.

After achieving a clinically complete response, she was followed-up regularly with serial chest CT scans that revealed only radiation pneumonitis until ten months following CRCT. In November 2001, a new cystic lesion with fat-like density on chest CT scans appeared in the previously irradiated site, and it slowly grew until October 2003. In addition, thickening of the right upper lobe bronchus became apparent on CT scans taken in June 2003, and another new low density mass replacing the right upper lobe appeared around the cystic mass lesion on CT scans in October 2003 (Fig. 3). The bronchoscopic biopsy specimen was histologically the same as

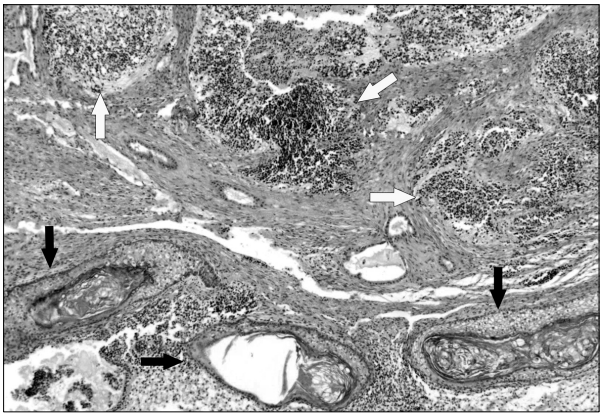

Fig. 1. Microscopic finding of immature teratoma of the nasal cavity: the tumor consisted of mature squamous cells with pilosebaceous differentiation (black arrow) and a neuroblastomatous component composed of compact sheets of small round cells (white arrow) with partial neuronal differentiation (H & E stain, magnification 40×).

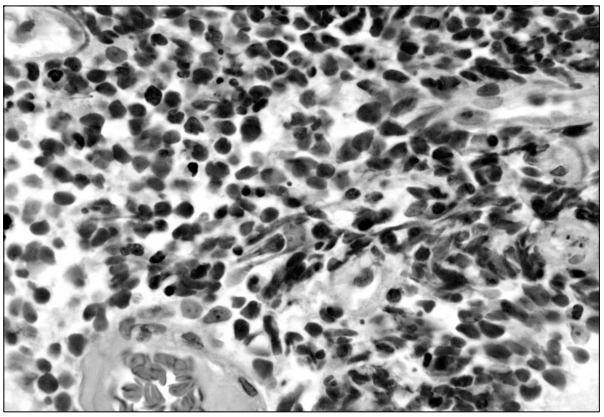

Fig. 2. Microscopic finding of bronchoscopic biopsy shows compact sheets of small round cells with hyperchromatic nuclei, inconspicuous nucleoli associated with squeezing artifacts and a scant amount of cytoplasm, suggestive of small cell carcinoma of the lung (H & E stain, magnification 400×).

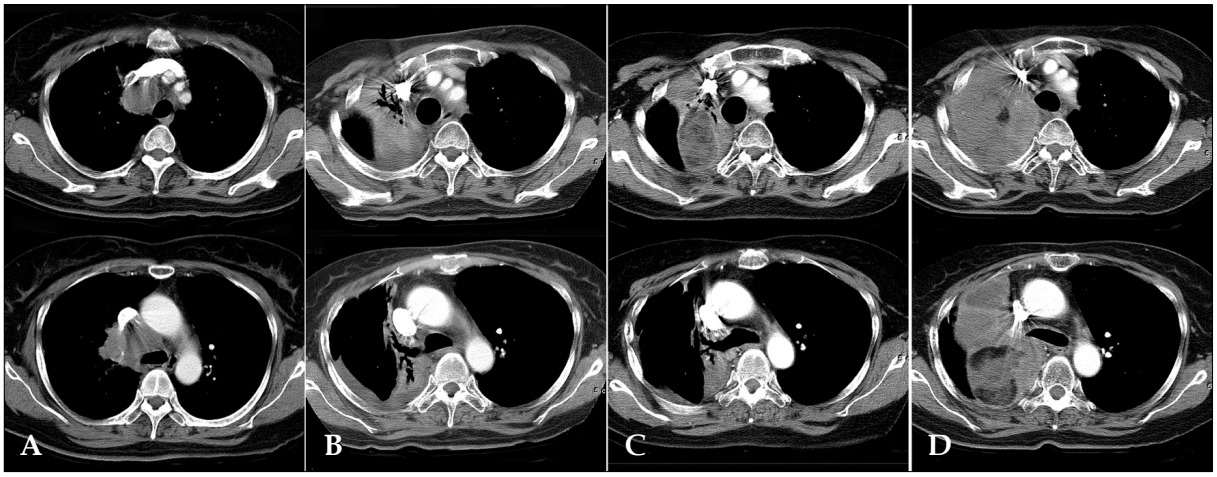

Fig. 3. Serial chest CT scans: (A) At the time of the small cell lung cancer diagnosis (Nov. '99); (B) Only radiation pneumonitis existed ten months after thoracic radiation therapy (Oct. '00); (C) A new mass with fat density was detected (Nov. '01); (D) A slow growing mass with fat density and another new mass replacing the right upper lobe (Oct. '03).

the initial bronchoscopic specimen, and there were increased uptakes in the right upper lung and right supraclavicular area on 18F-fluorodeoxyglucose positron emission tomography. Chemotherapy consisting of irinotecan and carboplatin for salvage purposes was begun, however, the patient died of septic shock two weeks later – 69 months after the initial diagnosis. Until her death, there was no evidence of tumor recurrence in the nasal cavity, nor elevation of teratoma markers throughout the disease course.

DISCUSSION

Sinonasal teratocarcinosarcoma (SNTCS), which was first described by Heffner et al.[1] in 1984, is uncommon and it might have variegated microscopic features of adenocarcinoma, rhabdomyosarcoma, and neuroblastoma. In addition, recurring tumors could also have variegated microscopic features.[1,4] Endo et al.[4] reported one SNTCS case with a history of three local recurrences. A patient with suspected rhabdomyosarcoma underwent a tumor resection after chemotherapy and the tumor recurred four years later. Resection plus postoperative radiation therapy was undertaken for the recurrent tumor, and thereafter two additional subtotal resections were carried out. The resected tumors consisted of epithelial, neuroblastic, and rhabdomyoblastic components. Cellular atypia and anaplasia of each component became more prominent in the recurrent tumors. Heffner et al.[1] reported on three SNTCS cases who developed cervical lymph node metastases that were pathologically confirmed. Two cases had singular histologic features (poorly differentiated adenocarcinoma or neuroblastomatous tissue) and one had a mixed and variegated appearance of poorly differentiated adenocarcinoma, poorly differentiated neuroblastoma, and cells with chondroid differentiation. Thus, it could be speculated that the secondary tumor with two different histologic types in the same patient treated for SNTCS could be a metastatic tumor rather than a new primary.

Non-germ cell malignant tumors arising from teratoma can metastasize.[3-6] But a metastatic tumor that was detected for the first time in a patient and has the histology of non-germ cell malignant tumor could be misdiagnosed as a metastatic disease of a non-germ cell tumor. Two other authors reported cases of misdiagnosis similar to our case.[2,3] Lim et al.[2] reported on a female patient who presented with a left supraclavicular lymph node enlargement. She was histologically diagnosed with a metastatic small cell carcinoma of an unknown origin. After nine courses of chemotherapy, an abdominal mass was palpated, which proved to be a small cell carcinoma of the pulmonary type arising in mature cystic teratoma of the ovary. Sekine et al.[3] reported on a male patient with a retroperitoneal mass arising 12 years after treatment of a testicular germ cell tumor. The histology of the retroperitoneal mass biopsy specimen was initially interpreted as adenocarcinoma, and they finally concluded that it represented the recurrence of a germ cell tumor for the following reasons: (1) the histology of the specimen was similar to an epithelial component of teratoma found in the tissue resected 12 years earlier; (2) a systemic survey failed to detect any other primary site; (3) the young age of this patient (35 years) was consistent with a germ cell tumor rather than an adenocarcinoma; and (4) the retroperitoneum is the most frequent site of late recurrences of testicular cancer.

The current case, which was treated for SNTCS, had a growing mass with fat-like density on serial chest CT scans after complete response of the small cell carcinoma treated as the second primary tumor. This phenomenon could be explained by the assumption that the sensitive small cell carcinoma component responded well to radiation therapy and chemotherapy while the resistant sarcomatous component grew slowly after finishing CRCT. This is analogous to the 'growing teratoma syndrome', which is the phenomenon whereby germ cell tumors enlarge after chemotherapy despite the complete eradication of malignant cells and the normalization of serum tumor markers, and it is caused by resistant non-germ cell malignancies.

The patient in this report presented with small cell lung cancer 21 months following the initial diagnosis of immature teratoma of the nasal

cavity. She was treated with CRCT as if it had been a primary small cell lung cancer and clinical complete response was achieved. Follow-up CT scans thereafter showed a mass slowly growing over two years, with fat-like density in the previously irradiated lung after the diagnosis of lung cancer. At the time of the small cell lung cancer diagnosis, we should have considered two possibilities of new second primary lung cancer and pulmonary metastasis from the neuroblastomatous component of the immature teratoma. Even though we failed to prove the presence of a lipomatous component in the lung specimen, we speculate that it was not a new primary small cell carcinoma of the lung, but that both the small undifferentiated cells and lipomatous components from the pluripotent germ cells had metastasized to the lung on the basis of the findings of serial chest CT scans. Another treatment option, which might have included surgical resection and/or chemotherapy of a different regimen to deal with the sarcomatous component, could have been considered on the condition that the possibility of metastasis from SNTCS had been suspected.

REFERENCES

1. Heffner DK, Hyams VJ. Teratocarcinosarcoma (malignant teratoma?) of the nasal cavity and paranasal sinuses A clinicopathologic study of 20 cases. Cancer 1984;53:2140-54.
2. Lim SC, Choi SJ, Suh CH. A case of small cell carcinoma arising in a mature cystic teratoma of the ovary. Pathol Int 1998;48:834-9.
3. Sekine I, Sasaki Y, Hasebe T, Mukai K, Kinoshita T, Amano A, et al. Recurrence of a germ cell tumor 12 years after initial treatment: a case report. Jpn J Clin Oncol 1998;28:50-3.
4. Endo H, Hirose T, Kuwamura KI, Sano T. Case report: Sinonasal teratocarcinosarcoma. Pathol Int 2001;51:107-12.
5. Ohtsuka M, Satoh H, Inoue M, Yazawa T, Yamashita YT, Sekizawa K, et al. Disseminated metastasis of neuroblastomatous component in immature mediastinal teratoma: a case report. Anticancer Res 2000;20: 527-30.
6. Motzer RJ, Amsterdam A, Prieto V, Sheinfeld J, Murty VV, Mazumdar M, et al. Teratoma with malignant transformation: diverse malignant histologies arising in men with germ cell tumors. J Urol 1998;159:133-8.