Case Report

Multiple Myeloma with Pleural Involvement after Pelvic Radiotherapy for Endometrial Carcinoma

Bülent Karagöz, Alpaslan Özgün, Oğuz Bilgi, Zafer Küçükkodacı, Alev Akyol Erikçi, Özkan Sayan, Emin Gökhan Kandemir

Abstract

Although ionizing radiation is strongest factor linked to multiple myeloma, increased myeloma risk has not been fully explained after pelvic radiation. Pleural involvement of MM is also rare. We present a MM case with pleural involvement as an unusual presentation diagnosed in fifth years of pelvic radiotherapy. A sixty-two-year-old woman with dyspnea and a mass in forehead was admitted to our clinic. Before five year, the patient had received pelvic external beam radiotherapy (RT) with dose of 40 Gy for endometrial adenocarcinoma. PET/CT scan detected FDG uptakes in frontal bone, right pleura, and sacrum. Lambda light chain type multiple myeloma with pleural involvement was diagnosed with histopathological examinations of frontal bone mass, bone marrow, and pleural fluid and with serum/urine electrophoresis. The patient died in second course of VAD chemotherapy. Although relation between increased myeloma risk and pelvic radiation is not clear and pleural involvement is rare, multiple myeloma should be included to the differential diagnosis in patients received pelvic radiotherapy or in unexplained pleural effusion.

Introduction

The leukemogenic and carcinogenic effects of ionizing radiation have long been recognized. Exposure of ionizing radiation is also strongest factor linked to an increased risk of multiple myeloma (MM) (1). A five times greater MM incidence has been reported in atomic bomb survivors (2). However, increased risk of MM has not been fully explained after pelvic radiation.

MM is malignant disease of bone marrow plasma cells with clonal proliferation. Pleural effusions occur in 6% of myeloma patients, but involvement of the pleura by MM is rare (3-5). We present a MM case with pleural involvement as an unusual presentation and five years after pelvic radiotherapy.

Case Report

A 62-year-old woman with dyspnea and a mass in forehead was admitted to our clinic. Before five year, the patient had received pelvic external beam radiotherapy (RT) with dose of 40 Gy for endometrial adenocarcinoma. Physical examination revealed right pleural effusion and a frontal calvarial 3 centimeters mass. In PET/CT imaging, 18F-FDG uptakes were detected in frontal bone, right pleura, and sacrum (Figure 1). Histopathological examination of frontal bone mass, bone marrow, and pleural fluid showed MM.

The nephelometric values of serum level of Ig A (0,17 g/L N: 0,82-4,53 g/L), IgM (0,05 g/L N:0,46-3,04 g/L), and IgG (3,35 g/L N:7,51-15,60 g/L) levels were decreased. In serum immunoelectrophoresis, IgG, IgA, IgM and kappa light chain serum levels were decreased lambda light chain level was normal. Urine electrophoresis showed that lambda light chain level was high.
With the present data, lambda light chain type MM with pleural involvement was diagnosed. After three times hemodialysis for acute renal failure, VAD chemotherapy regimen (vincristine 0.4 mg/day continuous infusion in 1-4 days, doxorubicin 9.0 mg/m²/day continuous infusion in 1-4 days and intravenous dexamethasone 40 mg/m²/day in 1-4, 9-12 and 17-20 days/4 weeks) was started. In first cycle, renal function returned to normal, dyspnea was decreased and frontal bone mass disappeared. The patient died in second cycle because febrile neutropenia.

Discussion

Several risk factors were described for myeloma risk. Hereditary and genetic factors may predispose to myeloma development. Various chemicals such as metals, especially nickel; agricultural chemicals; benzene and petroleum products; other aromatic hydrocarbons; agent orange; and silicon have been considered as potential risk factors. Retrospective cohort studies have established an association between MM and infectious or autoimmune disease. The strongest single factor linked to an increased risk of MM is ionizing radiation (1). After a latent period of approximately 20 years, a five times greater MM incidence has been reported in atomic bomb survivors (2). Unlike this documentation, risk of second myeloma after local radiation is controversial. Several studies did not suggest increased risk of MM after pelvic radiation. A retrospective study examined between pelvic RT and the development of subsequent hematological malignancies (6). There was no significant association between radiation and the development of multiple myeloma. In another study, the relationship between exposure to sparsely ionizing radiation and mortality due to cancers of hematopoietic and lymphopoietic tissues was studied in women treated by radiation for benign gynecological disorders (7). There was little or no evidence of effects attributable to radiotherapy for multiple myeloma (RR = 0.6; 90% CI 0.3-1.4).

However, a study showed increased risk of second malignant neoplasm outside radiation fields in patients with uterine cervical carcinoma, although radiation induced malignancies were expected locoregional (8). These authors reported a second myeloma after pelvic radiotherapy as well as breast, lung, bladder, thyroid cancer, and non-Hodgkin’s lymphoma. To the best of our knowledge, our presented patient is the second
case of MM after pelvic radiotherapy.

This case has also a rare presentation. Pleural effusions occur in multiple myeloma patient as well as thoracic skeletal lesions, intrathoracic plasmacytomas, and pulmonary paranchymal infiltrates. The causes of pleural effusion are nephrotic syndrome, pulmonary embolism, amyloidosis, secondary neoplasms and myeloma infiltration on the pleura, and lymphatic obstruction in multiple myeloma patients (3). Although pleural effusions occur in 6% of MM patients, pleural involvement of myeloma is rare (<1%) (4). In our case, plasmacytoma have diagnosed by biopsy of frontal mass and bone marrow examination showed multiple myeloma. Pleural fluid electrophoresis was not able to be performed due to her previous pleurodesis, but cytological analysis of pleural effusion specimen and FDG uptake in pleural mass suggest pleural involvement of myeloma.

Multiple myeloma should be included to the differential diagnosis in patients received pelvic radiotherapy or in unexplained pleural effusion.

References
1. Riedel DA, Pottern LM. The epidemiology of multiple myeloma. Hematol Oncol Clin North Am. 1992;6:225-47.
2. Ichimaru M, Ishimaru T, Mikami M, Matsunaga M. Multiple myeloma among atomic bomb survivors in Hiroshima and Nagasaki, 1950-76: relationship to radiation dose absorbed by marrow. J Natl Cancer Inst. 1982;69:323-28.
3. Alexandrakis MG, Passam FH, Kyriakou DS, Bouros D. Pleural effusions in hematologic malignancies. Chest. 2004;125:1546-55.
4. Manley R, Monteath J, Patton WN. Co- incidental presentation of IgA lambda multiple myeloma and pleural involvement with IgM kappa non-Hodgkin’s lymphoma. Clin Lab Haematol. 1999;21:61-63.
5. Sasser RL, Yam LT, Li CY. Myeloma with involvement of the serous cavities. Cytologic and immunochemical diagnosis and literature review. Acta Cytol. 1990;34:479-85.
6. Wright JD, St Clair CM, Deutsch I, Burke WM, Gorrochurn P, Sun X, Herzog TJ. Pelvic radiotherapy and the risk of secondary leukemia and multiple myeloma. Cancer. 2010;116:2486-92.
7. Inskip PD, Kleinerman RA, Stovall M, Cookfair DL, Hadjimichael O, Moloney WC, Monson RR, Thompson WD, Wactawski-Wende J, Wagoner JK. Leukemia, lymphoma, and multiple myeloma after pelvic radiotherapy for benign disease. Radiat Res. 1993;135:108-24.
8. Werner-Wasik M, Schmid CH, Bornstein LE, Madoc-Jones H. Increased risk of second malignant neoplasms outside radiation fields in patients with cervical carcinoma. Cancer. 1995;75:2281-85.