

Osteosarcoma in an adolescent previously treated for Hodgkin's Disease

Dear Editor,

Development of a secondary tumor is an ominous effect of primary cancer treatment.

An adolescent boy presented at his follow-up outpatient visit for Hodgkin's Disease (HD) with a painful swelling around his left knee. He was diagnosed with stage II HD 8 years before and treated with 8 cycles with ABVD (Adriamycin, Bleomycin, Vinblastine, Dacarbazine)¹. His family history was unremarkable for family cancer or cancer at young age. Diagnostic imaging studies including MRI, (coronal and axial image) confirmed an aggressive lesion of the distal femoral metaphysis with surrounding oedema and partial encasement of popliteal vessels. Technetium bone scan revealed fairly intense uptake at the site of the left distal femur without any metastasis or skip lesion. His chest CT scan was normal. A CT-guided biopsy confirmed the diagnosis of a localized high-grade osteosarcoma.

Due to his compromised cardiac function (LVEF of 48%), he was not eligible for doxorubicin administration, not even with the use with desrazoxane. Taken into consideration the rarity of the disease and the complexity of his treatment, we decided to start his neoadjuvant treatment according to the SFOP protocol, which excludes doxorubicin². After 10 weeks, he underwent surgical resection of the left distal femur and reconstruction with a limb salvage procedure. Histological examination of the resected tumour revealed good response to neoadjuvant chemotherapy with greater than 90% tumor necrosis. In total, he received 12 courses of High Dose-Methotrexate (12 g/m²) and 5 courses of Ifosfamide/Etoposide (500 mg/m² i.v / 14 g/m² i.v). At one-year follow-up, the chest CT scan revealed lung metastases for which he underwent successful metastasectomy.

Secondary malignancy is the second main cause of death after recurrence among childhood cancer survivors³. Although osteosarcoma is the most common second malignancy, only few reports regarding the efficacy of chemotherapy are available. Our patient was not exposed to irradiation and had a free family history of cancer, thus excluding p53 mutations, as in Li-Fraumeni syndrome. Nevertheless, he was previously exposed to alkylating agents (dacarbazine) as well as to anthracyclines, which are associated with increased risk of secondary osteosarcomas⁴.

This case report confers further evidence that treatment for secondary osteosarcoma can be accomplished according to the guidelines for primary tumor modified for previous drug exposure. Long-term surveillance is considered mandatory. Treatment for relapse within the lungs is primarily surgical and complete removal of all metastases should be attempted. Even patients with multiple recurrences may be cured as long as recurrences are resectable, and repeated thoracotomies are often justified⁵.

Results from multicenter collaborative studies which address the issue of therapy for secondary osteosarcoma are anticipated in order to create a standard for the management of secondary osteosarcoma, as well as to assess the safety and efficacy of treatment regimens.

Conflict of interest

None declared.

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Kourti M, Sidi V, Papakonstantinou E

Pediatric Oncology Department, Hippokratia General Hospital, Thessaloniki, Greece

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Corresponding Author: Maria Kourti, Sakellaridi 25, 54248, Thessaloniki, Greece, tel/fax: +302310329729, e-mail: makourti@med.auth.gr