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Cutaneous Ewing's sarcoma secondary to chemotherapy given for testis tumor: Case report

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ABSTRACT

INTRODUCTION: Testicular cancer has high cure rates, especially after the adjuvant use of chemotherapy. Secondary tumors may develop months and years after the primary tumor. We aimed to report a case of cutaneous Ewing's sarcoma at the site of surgery 3 years after BEP chemotherapy.
PRESENTATION OF CASE: 21 year old male underwent radical orchiectomy in 2008. After one year surgical site complaints brought him to same hospital. A limited surgical resection was made. As his complaints continued he applied to our clinic. We resected the lesion with a 5 cm safety margin with the light of previous medical history. Pathology revealed cutaneous Ewing's sarcoma, and patient received VACD-IE chemotherapy. He is free of recurrence till now.
DISCUSSION: Chemotherapy may cause secondary cancer especially in long term. In this case secondary tumor is diagnosed three years after surgery. Patient underwent therapeutic surgery and received chemotherapy (VACD-IE) for secondary Ewing's sarcoma. Early diagnosis and definitive treatment provide recurrence free survival in the patient.
CONCLUSION: Secondary tumors can emerge months or years after primary tumor therapies, and are not related with the primary tumors. Any lesion or sign should be investigated carefully. Early diagnosis and correct treatment could prevent dramatic results.

1. Introduction

Testicular cancer is the most common solid tumor among males in the second and third decade of life.[1] Testicular cancer has one of the highest cure rates of all cancers: a five-year survival rate in excess of 90 percent overall.[2] Treatment options of testis tumor depend on the stage of cancer, patients' tolerability, and the experience of the physician. BEP chemotherapy, consists of bleomycin, etoposide, and cisplatin, is the choice of treatment in metastatic testicular germ cell tumors. Tumors developed after radiotherapy or chemotherapy are called secondary tumors. Secondary tumors are independent of primary tumors, and can arise months and years after the primary tumor.[3]

We aimed to report a case with cutaneous Ewing sarcoma at the site of surgery 3 years after BEP chemotherapy. To our knowledge it is the first case of secondary Ewing's sarcoma after chemotherapy of testicular cancer.

2. Presentation of case

21 year old male patient underwent radical orchiectomy for testicular mass in a state hospital in 2008. The patient had no previous story of familial testicular cancer and other malignancies. Pathology report revealed mixed germ cell tumor, and patient received 2 cycles of BEP chemotherapy in the same hospital. The patient presented a year later with discharge and inflammation at the site of the wound. A limited surgical resection was performed and pathology revealed atypical cellular differentiation and foreign body reaction. Patient's complaints continued after this operation and referred to our clinic after 12 months in 2011 (Fig. 1). Laboratory investigations including complete blood count, blood urea nitrogen, creatinine, alpha feto protein, β human chorionic gonadotropin, carcinoembryogenic antigen, and lactate dehydrogenase levels were in normal ranges. Ultrasound reported that the lesion was limited to skin and subcutaneous tissue. We resected

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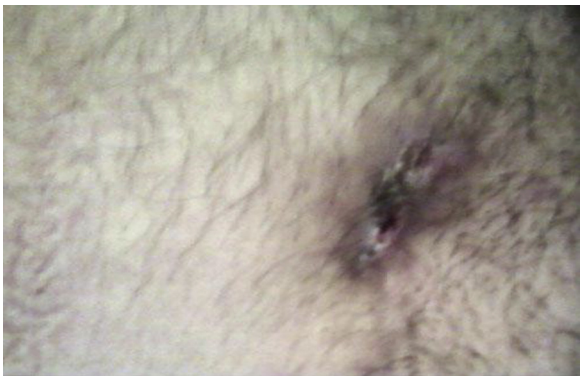

Fig. 1. The gross appearance of cutaneous lesion at the site of surgery.

the lesion with a 5 cm safety margin with the light of previous medical history. Pathology revealed cutaneous Ewing sarcoma with negative margins (Fig. 2). The patient had received vincristine, actinomycin D, cyclophosphamide, doxorubicine, ifosphamide and etoposide (VACD-IE) chemotherapy regime and is free of recurrence till September 2013.

3. Discussion

Secondary tumors can emerge months or years after primary tumor therapies, and are not related with the primary tumors. The chemotherapeutics used for the primary tumor, inactivate the cancer cells by disrupting cellular division, and may also affect normal cells in many ways.[3] Secondary tumors have different characteristics than primary tumors even in the same region. Furthermore primary therapy regimes could restrict secondary therapy, and reduce survival.[4,5]

Travis et al. studied 40,576 testicular cancer patients with at least one year of survival. They reported 2200 secondary solid tumors in these patients. This study demonstrated the increased incidence in secondary tumors such as leukemia and some solid tumors (lung, thyroid, esophagus, stomach, pancreas, colon, rectum, kidney, bladder, connective tissue cancers and malignant mesothelioma). They also reported that younger patients at the time of diagnosis might experience high risk of secondary tumors as they get older.[6]

A similar study was conducted in Holland, reported subdiaphragmatic radiotherapy increases the risk of secondary tumors. The study also demonstrated a 4% increased risk in radiotherapy and 1.5% increased risk in chemotherapy for secondary tumors in 20 years of follow-up.[7]

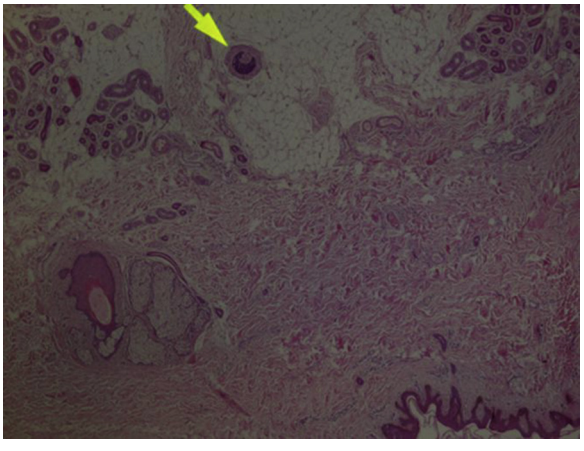

Fig. 2. Cutaneous metastasis of Ewing's sarcoma. The vascular invasion of metastatic tumor cells (arrow) (Hematoxylin and Eosin stain 40×).

Secondary cancers can occur following treatment with chemotherapy. Cisplatin as an alkylating agent and etoposide as a Topoisomerase-II inhibitor have been shown to be the cause of secondary cancers.[8] It was also shown that etoposide and cisplatin had synergistic effects for secondary leukemia when used together for chemotherapy.[9]

The standard chemotherapy of Ewing's sarcoma has been evolved since the first independent reports of Sutow, and Pinkel.[10,11] In 1974 Rosen et al. reported first results of radiotherapy with four drug regimen consisting of vincristine, actinomycin D, cyclophosphamide, and doxorubicin (VACD) leading to long term survival.[12] The Pediatric Oncology Group–Children's Cancer Group (POG-CCG) reported a comparative study of VACD versus VACD plus etoposide and ifosphamide (IE). VACD-IE group achieved a 5 year event-free survival (EFS) rate of 69% versus 54% in the VACD group.[13]

Chemotherapy may cause secondary cancer especially in long term. In this case secondary tumor is diagnosed three years after surgery. Patient underwent therapeutic surgery and received chemotherapy (VACD-IE) for secondary Ewing's sarcoma. Early diagnosis and definitive treatment provide recurrence free survival in the patient.

4. Conclusion

Secondary tumors may arise years after chemotherapy. Any lesion or sign should be investigated carefully. Early diagnosis and correct treatment could prevent dramatic results.

Conflict of interest

We certify that there is no conflict of interest with any financial organization regarding the material discussed in the article.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and case series and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Serhat Tanik performed the orchiectomy. Kursad Zengin and Sebahattin Albayrak wrote the manuscript. Necip Pirincci and Recep Eryılmaz performed the last operation and chemotherapy. Deniz Yilmaz performed the pathological evaluation.

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