

Testicular Involvement in Relapsed Hodgkin Lymphoma

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Dear Editor,

Malignant lymphoma of the testis generates 5% of all testicular malignancy and 1% of all lymphomas (1). Testicular involvement is an extremely rare presentation of Hodgkin's lymphoma (HL); to date, testicular involvement has been reported in 5 patients with HL. We have examined a patient with testicular involvement of HL who relapsed 11 years after treatment. Written informed consent was obtained from patient.

A 40-year-old male patient was admitted to hospital in June 2014 with complaints of fever, weight loss and night sweats for 2 months. We found that he had been diagnosed as mixed cellularity classical HL (stage 2A) in 2003 and achieved complete remission after 3 cycles of Adriamycin (Doxorubicin,

Koçak; Tekirdağ, Turkey), bleomycin (Blemisin, Koçak; Tekirdağ, Turkey), vinblastine (Vinko, Koçak; Tekirdağ, Turkey), dacarbazine (Dakarbaz, Koçak; Tekirdağ, Turkey) (ABVD) chemotherapy and involved field radiotherapy. He was followed without any therapy for 11 years. On admission, physical examination was unremarkable except for millimeter cervical lymph nodes. The following measurements were recorded: erythrocyte sedimentation rate of 97 mm/h, lactate dehydrogenase level 247 U/L and beta2-microglobulin of 2.14 mg/L. 18F-fluorodeoxyglucose (FDG)-positron emission tomography (PET-CT) (Siemens; Munich, Germany) revealed multiple cervical, mediastinal and abdominal lymph nodes with increased FDG uptake. In addition, right (SUV_{max}: 20.16)

TABLE 1. Clinical characteristics, pathological features and treatment of patients with testicular Hodgkin's lymphoma in the literature (3).

| Author | Age | Clinical information | Site (testis) | HL subtype | IHC | Stage | Treatment | Outcome |
|-------------------------------------|-----|---|---------------|---------------------------|----------|-------|-------------------------------------|-----------------------|
| Our case | 40 | Cervical, mediastinal, abdominal mass, testicular involvement (relapse) | Bilateral | MC | CD15 (+) | IVB | CT (6xABVD) | CR (DFS 1 years) |
| Ben Ameer El Youbi and Errihani (3) | 17 | Cervical mass, Empty scrotum (initial involvement) | Left | NS | CD15 (+) | | | |
| | | | | | CD30 (+) | IVB | Orchiectomy + CT (8xABVD) | CR (DFS 2 years) |
| Gatt et al. (1) | 73 | Testicular mass (initial involvement) | Right | NS | CD3 (+) | IVA | Orchiectomy (Not taken CT) | Died before CT |
| | | | | | CD15 (+) | | | |
| | | | | | CD30 (+) | | | |
| Seliem et al. (2) | 52 | Testicular mass (initial involvement) | Left | Classical | CD15 (+) | IVB | Orchiectomy + CT (4xABVD and 2xAVD) | CR (Not reached) |
| | | | | | CD30 (+) | | | |
| Glaholm et al. (4) | 56 | Scrotal pain (initial involvement), Axillary node | Right | MC | np | IVA | Orchiectomy + CT (6xChlVPP) | CR (DFS 3 years) |
| Vishniavsky et al. (5) | 79 | Inguinal mass (initial involvement) | Right | Hodgkin sarcoma (autopsy) | np | - | - | Died before treatment |

HL: Hodgkin's lymphoma; IHC: immunohistochemistry; NS: nodular sclerosis; MC: mixed cellularity; np: not performed; CT: chemotherapy; ABVD: doxorubicin, bleomycin, vinblastine, dacarbazine; AVD: doxorubicin, vinblastine, dacarbazine; ChlVPP: chlorambucil, vinblastine procarbazine, prednisolone; CR: complete response; DFS: disease free survival.

This study has been presented at the 41st National Hematology Congress, November 2015, Antalya, Turkey

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and left (SUV_{max} : 19.95) testicular involvement was observed by PET-CT. Scrotal ultrasound showed a slightly heterogeneous left testicular parenchyma without any mass. The diagnosis of mixed cellularity classical Hodgkin lymphoma was performed with excisional biopsy from mediastinal 41x29 mm conglomerate lymphadenopathy. In addition, CD30 (+) and CD15 (+) were histologically positive. Stage 4B Hodgkin's lymphoma has been confirmed because of the testicular involvement and the patient has been treated with 6 cycles of ABVD. PET-CT after chemotherapy showed complete metabolic remission.

Testicular involvement of Hodgkin lymphoma is a rare condition. So far, five cases have been reported in the literature. Along with our case, features of a total of 6 cases are shown in Table 1 (1-5). Half of the patients were admitted with testicular mass; however, there were no testicular masses reported in the other patients, or in our case. Unilateral testicular involvement was present in other cases, while bilateral involvement was detected in our case. We did not perform orchiectomy because of the increased FDG uptake in other areas of lymphadenopathies, whereas orchiectomy was performed in other cases. Following chemotherapy, the disappearance of testicular involvement supported our diagnosis. Another feature that distinguishes our case from the others was the presence of testicular involvement in the recurrence of HL; other patients were admitted with testicular mass or involvement at first diagnosis.

Testicular involvement may be due to the directly contiguous spread from adjacent lymph nodes or hematogenous spread (2). In our case, we can assume that testicular involvement is spread hematogenously because there was no lymph node involvement in the adjacent testes. There is no definitive treatment recommendation because of the small number of cases, and ABVD chemotherapy remains a standard treatment in HL. There are also no precise data about the necessity of orchiectomy or central nervous system (CNS) prophylaxis.

As a result, testicular involvement is rare in HL, but should be considered, and orchiectomy should be avoided. There is no definitive information about the prognostic significance of

the testicular involvement of Hodgkin's lymphoma because of the inadequate number of these patients and short follow-up times. In the future, we hope to obtain evidence-based information about CNS prophylaxis and the duration of treatment by increasing the number of cases analyzed.

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