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CASE REPORT

Primary testicular lymphoma: Two case reports and review of the literature

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Summary ***Primary testicular lymphoma, is a rare testis tumor that accounts for only less than 9% of all testis tumors. In the preoperative period, it is extremely difficult to distinguish this tumor from other testis tumors. Its diagnosis is done by histological analysis. Most commonly encountered histological type is diffuse large B-cell lymphoma. Adjuvant radiotheraphy and/or chemotheraphy is given after orchiectomy. Prognosis is worse than other testis tumors. Non-metastatic tumors indicates good prognosis within one year. Ongoing research in patients with primary testicular lymphoma, are on efficacy of adjuvant theraphies and preventive and cure effect on extranodal extension to central nervous system which is the most common site for recurrency. There are conflicting results because of the small number of patient size. Here we present two cases with primary testicular lymphoma at the ages 71 and 82.***

KEY WORDS: *Primary testicular lymphoma; Orchiectomy; Chemotheraphy; Radiotheraphy.*

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INTRODUCTION

Primary testicular lymphoma (PTL) is an extranodal lymphoma in which primary origin is testis, and it accounts only 1-2% of all non-Hodgkin lymphomas (NHL) and 1-9% of all testis tumors (1, 2). Most of the patients are above 60 years (3) and PTL is the most commonly seen neoplasm in this age group (4).
Here we present two cases with primary testicular lymphoma.

CASE REPORTS

Case 1

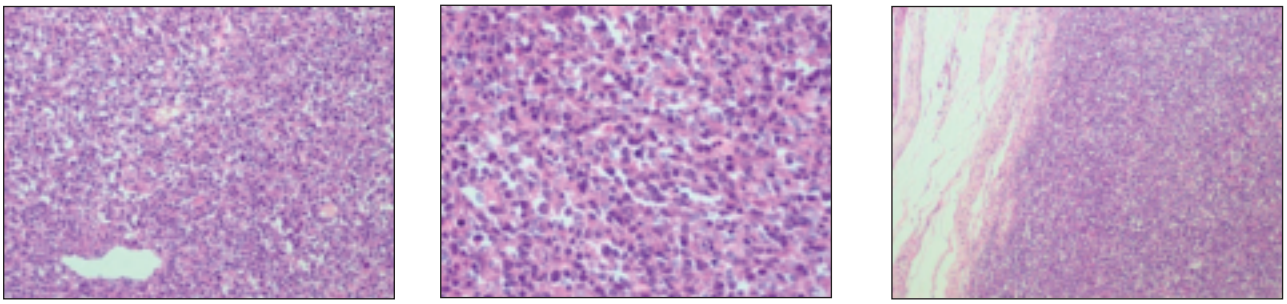
A 70 year-old male patient, was admitted to the urology outpatient clinic with rapid onset painful swelling on his right testis. On physical examination, right scrotal mass palpation with erythematous scrotum, immobile testis, decreased fluctuation and Prehn's sign indicating orchitis were observed.
At diagnostic with scrotal ultrasonography (USG), there was diffuse enlargement and increased vascularisation of the right testis when compared with the left one. Testis parenchyma was reported as isoechoic.
There was no peripheric lymphadenopathy (LAP) or hepatosplenomegaly. Laboratory examinations showed hemoglobin 13.57 gr/dL (MCV: 86.61 fL, MCH: 31 pg, MCHC: 36.27 g/dL); blood leucocyte: 4500/mm^3 (65% neutrophil, 35% lymphocyte); trombocytes: 210000/mm^3; sedimentation rate: 5/h; serum reactive protein: 0,35 ng/ml; LDH: 212 UI/L (100-190); SGOT:35; SGPT: 42; GGT: 38 U/L. Tumor markers were negative: alpha-fetoprotein (AFP) 1,1 U/L and beta human chorionic globulin (HCG) 0.05 mU/ml. He was diagnosed as orchitis and given 2 week antibiotic and antiinflammatory treatment. At his control visit after treatment, acute symptoms as scrotal pain, testicular pain, and erythema were dissappeared. However, testis enlargement persisted and scrotal Doppler USG was performed again. There was reactive hydrocele, the right testis was larger than the left one with no vascularization difference and orchiectomy was planned. Right high orchiectomy was performed. At histopathological evaluation, the cells had large nucleus and some of them had prominent nucleolus. AT immunohistochemistry analysis the neoplastic cells were CD20 and bcl2 positive; Bcl6, CD30 and ALK negative. Ki67 positivity was above 80% in the areas with good fixing. non-Hodgkin diffuse large B-cell lymphoma diagnosis was made (Figure 1).
The abdominopelvic, thorax and cranial CT were normal. He was in stage I and adjuvant chemotheraphy was planned, 6 dose of cyclophosphamid, novantron, oncovin, prednisolone (CNOP) chemo-treatment was done. There was no pathology in abdominal and thorax CT, andno other accompanying pathology at 3 year follow up.

Case 2

An 82 year-old male patient was admitted to the urology outpatient clinic with fatigue for 3 months, back pain, painless swelling in his left testis. On physical examina-

No conflict of interest declared.

Figure 1.
Diffuse lymphoma infiltration in the interstitial region between seminiferious tubules in the testis (HE x 100).



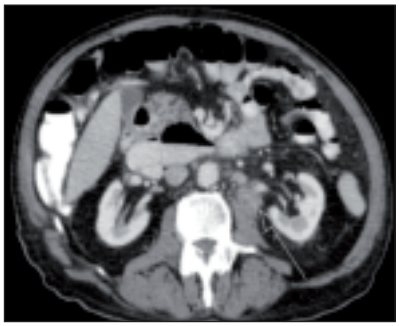


Figure 2a.
26 mm diameter mass lesion with irregular contours at left renal hilus level in the retroperitoneal area.

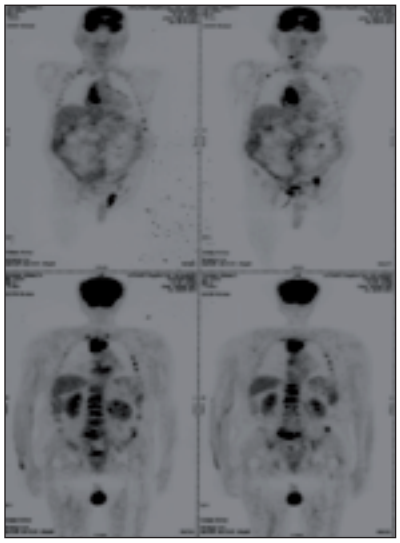


Figure 2b.
Multiple hypermetabolic metastatic lesions in bilateral cervical lymph node stations, lung parenchymal and mediastinal regions, abnominopelvic organs and lymphatic nodes and skeletal system.

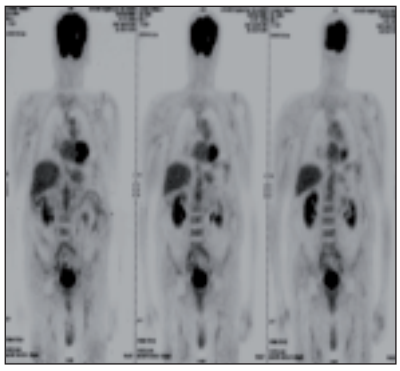


Figure 2c.
Complete metabolic/morphologic regression with absence of hypermetabolic lesions in organs and lymphatic nodes.

tion, he was in normal general condition, well oriented and cooperative, with good cognitive status. Blood pressure was 110/60 mm Hg, pulse 76/minute,temperature 36,1 0C, costovertebral angle tenderness (CVAT) -/-, suprapubic tenderness was positive and there was no bladder overdistension. The right testis was in the scrotum with normal dimensions, and epididymis also palpated normally. There was a big and hard mass of the left testis. On digital rectal examination a +1 fibroadenoma texture of prostate was found.

At diagnostic ultrasound, there was a diffuse enlarged and vascularized left testis when compared with the right side and testis parenchyme was isoechoic.

At physical examination there was no peripheric LAP or splenomegaly. His laboratory investigation results were as follows: hemoglobin: 11,3 gr/dL (MCV 83,8 fL, MCH 27,8 pg, MCHC 33,1 g/dL); blood leucocytes: 6760/ mm^3 (65% neutrophil, 35% lymphocyte); thrombocytes: 225 000/mm^3; sedimentation rate: 81/h; serum reactive protein: 14 ng/ml; LDH: 443 UI/L (135-225), SGOT: 21.8; SGPT: 7.07 U/L. Tumor markers were negative: alpha-fetoprotein (AFP) 2 ng/mL (< 7) and beta human chorionic globulin (HCG) 0,1 mU/mL (< 2). Right inguinal orchiectomy was performed. At pathologic examinations there were diffusely scattered, prominently large, pleomorphic nuclei with irregular contour and thin chromatin, some cells with multilobulated and prominent nucleoulus, wide cytoplasmic cell neoplastic lymphoid infiltration in testis tissue and spermatic cord. The epididymis and rete testis tissue showed normal morphologic appearance. At immunohistochemistry the cells forming lesion were found CD20 (+) and CD3 (-). For typization a wide histochemistry analysis was necessary. The neoplastic cells were CD20 and bcl2 positive; Bcl6, CD30 and ALK were negative; Ki67 positivity was above 80% in the areas with good establishment. The patient was diagnosed with non-Hodgkin high grade diffuse large B-cell lymphoma.

At abdominopelvic tomography (CT) there was a 26 mm diameter mass lesion with irregular contours at left renal hilus level in the retroperitoneal area. The lesion was adherent to the anterior surface of the psoas muscle. The lower segmentary branch of the left renal artery was surrounded by the mass lesion. There were multiple lymphadenopathies in the left paraaortic region with maximum size 23 x 18 mm, edema in the extraperitoneal region, andlymphadenopathies localized at interaortacaval, retrocaval, bilateral common iliac, external and internal iliac chain, with maximum diameter of 12 mm (lymphoproliferative diseases?/metastasis?) (Figure 2a).

The thorax and cranial CT were normal. The initial staging fluorodeoxyglucose (FDG) PET examination showed a maximum 34 mm large LAP with hypermetabolic activity and increased uptake, pericardial inclusion, LAP with hypermetabolic activity at left renal hilus level and at internal and external iliac level. Mass lesion with hypermetabolic activity at the left testis of 65 x 33 mm dimension and multiple metastasis lesions at skeletal system were observed (Figure 2b).

According to those findings, the patient was classified as stage IV and a 6 dose rituximab, cyclofosfamide, epirubicin, vincristine, prednisolone, zoledronic acid with a 2 dose maintenance rituximab therapy was planned. After the 4th dose, FDG PET was performed to evaluate the response. There was total metabolic and morphologic response so the treatment protocol was continiued. There was no pathology at abdominal CT after six doses. At the end of the maintanance therapy, FDG PET was performed again.

Total metabolic and morphologic response was confirmed (Figure 2c). At the second year follow up, there was a right axillary LAP at chest CT with a ground glass density nodule near to the pleura of the left lung, a mass lesion of theright surrenal gland and thickening of the left side.

Written consent was obtained from the patient and their relatives for publication of the study.

Conclusions

In conclusion, as PTL is a rare disease, there is lack of data that can guide the treatment. However with the aid of retrospective data evaluation, better prognosis was obtained for nodal lymphoma. Despite the improvements in local and systemic disease central nervous system (CNS) relapse remains the worst complication.. Strategies that may decrease the risk of PTL patients will end up with better prognosis.

Introduction, Discussion and Supplementary References are posted as Supplementary Materials on www.aiua.it

References

1. Freeman C, Berg JW, Cutler SJ. Occurrence and prognosis of extra nodal lymphomas. Cancer. 1972; 29:252-60.

2. Shahab N, Doll DC. Testicular lymphoma. Semin Oncol. 1999; 26:259-69.

3. Sussman EB, Hajdu SI, Lieberman PH, Whitmore WF. Malignant lymphoma of the testis: a clinicopathologic study of 37 cases. J Urol. 1977; 118:1004-7.

4. Zucca E, Conconi A, Mughal TI, et al. Patterns of out come and prognostic factors in primary large-cell lymphoma of the testis in a survey by the International Extranodal Lymphoma Study Group. J Clin Oncol. 2003; 21:20-7.

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