First Case Report of Primary Testicular Diffuse Large B-Cell Lymphoma from the Western Region of Saudi Arabia

Abdulkader Albasri1*, Mohammed A. Elkablawy1,2, Ahmed Abdelbadie1, Akbar S. Hussainy1, Abdelaziz R. Aljohani3

1Department of Pathology, College of Medicine, Taibah University, Al-Madinah Al-Munawarah, KSA
2Department of Pathology, Faculty of Medicine, Menoufia University, Menoufia, Egypt
3Department of Pathology, King Fahd Hospital, Al-Madinah Al-Munawarah, KSA

ABSTRACT
Primary testicular lymphoma (PTL) represents 1-2% of all types of non-Hodgkin lymphomas (NHLs) and 1-10% of testicular neoplasms. Up to the best of my knowledge, this is the first case of PTL of the diffuse large B-cell lymphoma (DLBCL) in a 60-year-old man presented with a painless mass in the left testis as revealed by physical examination in a tertiary care hospital in Al-Madinah Al-Munawarah in the western region of the Kingdom of Saudi Arabia (KSA). Radiological examination revealed a large well-defined heterogeneous predominantly hypo-echoic mass with increased vascularity in the upper portion of the testis. On the other hand, histopathological examination revealed a tumor involving the whole left testis, which was large (measuring 6 × 3.5 × 3.3 cm), solid and dark red with focal areas of hemorrhage and epididymal infiltration. Immunohistochemistry showed positivity of leucocyte common antigen (LCA), pan B-cell marker (CD20) and negativity of pan T-cell marker (CD3). Other immunohistochemical markers such as CD10, placental alkaline phosphatase (PLAP), cytokeratin, vimentin, desmin and S100 protein were also negative. However, there was a marked expression of Ki67 and Bcl2 markers. Accordingly, the diagnosis of DLBCL was established. The tumor was classified as stage I according to the Ann Arbor system. The case was treated by orchectomy followed by prophylactic anthracycline-based chemotherapy and irradiation of the contralateral testis and central nervous system.

Keywords: Primary testicular lymphoma, Diffuse large B-cell lymphoma, Testis, Al-Madinah Al-Munawarah, Saudi Arabia

*Corresponding author: A. Albasri (abdbasri@hotmail.com)
1. Introduction

Primary testicular lymphoma (PTL) represents 1-2% of all types of non-Hodgkin lymphomas (NHLs) and 1-10% of testicular neoplasms affecting the elderly with a poor prognosis. The most common histological variant of PTL is the diffuse large B-cell lymphoma (DLBCL). Although DLBCL is the most common histopathological type of PTL, secondary infiltration of the testis, especially in high-grade Burkitt’s lymphoma, is more prevalent. The most common clinical presentation of PTL is a unilateral testicular mass. No case report or case series of PTL have been reported from the western region of the Kingdom of Saudi Arabia (KSA), and the present case is the first PTL of the DLBCL type to be reported from the western region of the KSA.

2. Case presentation

A 60-year-old man presented at the Urology Department of a tertiary care hospital in Al-Madinah Al-Munawarah in the western region of KSA in October 2017. He complained of a painless swelling in his left testis for three weeks, which was revealed as a firm mass by physical examination. Besides the absence of systemic symptoms, neither regional/generalized lymphadenopathy nor hepatosplenomegaly was detected. In addition, head, neck, chest and abdomen investigations were normal. Hematological investigations revealed a hemoglobin level of 14.4 g/dl, a total leucocyte count of 9.8×10^9/L, a platelet count of 250×10^9/L. Biochemical investigations revealed normal renal function tests and liver enzyme profiles. Ultrasonography revealed a large (2.7 × 1.7 cm) well-defined heterogeneous and predominantly hypo-echoic mass with increased vascularity in the upper portion of the left testis. Left testis measured 4.8 × 2.8 × 2.4 cm, while the right testis was normal except for a small epididymal cyst measuring 1.5 × 0.8 cm. Computed tomography scan of the abdomen and thorax revealed no abnormalities.

After undergoing left inguinal orchiectomy, histopathological examination revealed a tumor involving the whole left testis. The tumor was solid and dark red with focal areas of hemorrhage and measuring 6 × 3.5 × 3.3 cm. The tumor was infiltrating the epididymis but without infiltration of the tunica albuginea or tunica vaginalis. The relevant histopathological microphotographs are shown in Figure 1 (A-C).

![Figure 1](image-url)

**Figure 1.** Testicular invasion by large B-cell lymphoma at different magnifications: (A) 100x, 100 μm scale bar; (B) 200x, 50 μm scale bar; (C) 400x, 25 μm scale bar.
Immunohistochemistry showed positivity of leucocyte common antigen (LCA), pan B-cell marker (CD20) and negativity of pan T-cell marker (CD3). Other immunohistochemical markers such as CD10, placental alkaline phosphatase (PLAP), cytokeratin, vimentin, desmin and S100 protein were also negative. However, there was a marked expression of Ki67 and Bcl2 markers. Accordingly, the diagnosis of DLBCL was established. The tumor was classified as stage I according to the Ann Arbor system.\(^{(3)}\)

3. Discussion

Up to the best of my knowledge, no individual case or case series of PTL have been reported from the western region of KSA. However, only a single case of PTL was documented out of 19 cases of testicular cancers among patients from Dammam in the eastern region of KSA and presented at the 22\(^{nd}\) Saudi Urological Conference in March 2010.\(^{(4)}\) PTL represents up to 10\% of all testicular cancers.\(^{(1,5)}\) Moreover, DLBCL is the most common type of PTL, representing 69–90\% of PTL.\(^{(5)}\) Approximately 60\% and 30\% of patients are in stage I and II, respectively, and bilateral testicular involvement can be found in approximately 35\% of cases.\(^{(5)}\) In accordance with the published literature, the present case was in the seventh decade of life and presented with unilateral painless testicular mass but without B symptoms.\(^{(6)}\)

According to the Ann Arbor staging system,\(^{(3)}\) the PTL of the present case is classified as stage I. Post-operatively, the patient received anthracycline-based chemotherapy and irradiation of the contralateral testis and central nervous system. Complete remission was achieved with no serious side effects of the chemoradiotherapy. The patient was on follow-up with no recurrence or relapse after three years. Therefore, the same pattern of treatment was given to the patient, where the patient’s follow-up revealed no recurrence or relapse.

No standard treatment modality exists for PTL because it is a rare incidence. However, R-CHOP (rituximab, cyclophosphamide, doxorubicin, vincristine and prednisone) are used for the treatment of stage I and II PTL, and prophylactic scrotal radiotherapy together with systemic chemotherapy are performed after orchiectomy.\(^{(7)}\) Distant relapses often occur in most patients with stage I PTL if adjuvant therapy is not administered after orchiectomy. In stage II, radiotherapy of localized lymph nodes can prevent nodal relapses. In stages III-IV, systemic and intrathecal chemotherapy as well as scrotal radiotherapy are performed.\(^{(8)}\)

4. Conclusions

This is the first report of DLBCL type of PTL from Al-Madinah Al-Munawarah in the western region of KSA. PTL is a rare tumor of the testes but can be the most common testicular tumor in the elderly. Although PTL is a disease with a poor prognosis, no recurrence or relapse was observed in the present case on follow-up for more than three years. Because of the rare incidence of PTL besides its different development and progress in comparison to germ-cell testicular cancers, it is difficult to determine the treatment procedures to be applied after orchiectomy. Therefore, PTL should be considered as a manifestation of systemic diseases of the testis. Moreover, contralateral testis and relapse in the central nervous system should always be considered. PTL should also be considered in patients presenting with a painless mass in the testis, where a joint action should be undertaken by the urologist, pathologist, and oncologist investigating the case. We have reported the first case of PTL from Al-Madinah Al-Munawarah in the western region of KSA.
Ethical considerations

The present study was approved by the Ethics Committee of King Fahd Hospital, Al-Madinah Al-Munawarah - KSA. Written informed consent for publication of this case report was obtained from the patient.

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Authors’ contributions

A.A. designed the case; M.A.E. collected the data; A.A. conducted data interpretation; A.S.H. drafted the manuscript; A.R.A. revised the manuscript. All authors approved the final submission.

Competing interests

The authors declare that they have no competing interests associated with this article.

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