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CASE REPORT

Primary Lymphoma of the Testis in Remission for more than Ten Years: A Case Report

Lymphome primaire du testicule en rémission depuis plus de dix ans: Un rapport de cas

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ABSTRACT

BACKGROUND: **Primary testicular lymphoma is a unique, rare and aggressive extra nodal non-Hodgkin's Lymphoma (NHL). It is the most common testicular tumour in males between 60 and 80 years old**

OBJECTIVE: **To report a case of primary testicular lymphoma in a young man who has done very well on surgery and chemotherapy.**

METHODS: **The patient a Nigerian male civil servant who was single and aged 31 years presented to us with a history of a progressive painful right scrotal swelling of two years duration and associated painful right groin swelling for one year. A working diagnosis of right hydrocele with differential diagnosis of lymphangioma was made. Invesigations were essentially normal and patient was scheduled for hydrocelectomy but findings at operations were different.**

RESULTS: **At surgery, there was grossly enlarged hard right testis with thickened spermatic cord. Right radical inguinal orchidectomy with excision of the right spermatic cord and a regional lymph node was carried out. Histology revealed intermediate grade nodular NHL of the right testis. He was treated with systemic combination chemotherapy and has since been in complete remission for over 10 years.**

CONCLUSION: **Primary testicular lymphoma is a rare and unique neoplasm of the testis and is most commonly seen in men over the age of 60, but should be considered in the differential diagnosis of testicular tumours in younger age groups. It is curable in early stage with appropriate multimodalities of management. WAJM 2009; 28(6): 388–390.**

Keywords: **Lymphoma, testis, primary, remission.**

RÉSUMÉ

CONTEXTE: *lymphome testiculaire primaire est un appoint non unique, rare et agressive nodale lymphome hodgkinien (LNH). Elle est la tumeur la plus fréquente des testicules chez les hommes entre 60 et 80 ans*

OBJECTIF: *Rapporter un cas de lymphome testiculaire primaire d'un jeune homme qui a très bien fait sur la chirurgie et la chimiothérapie.*

Méthodes: Les patients un domestique civile nigériane qui était célibataire et âgé de 31 ans, présentés à nous avec une histoire d'un scrotum progressive droit gonflement douloureux d'une durée de deux ans et associée à l'aine droite gonflement douloureux pendant un an. Un diagnostic de travail de l'hydrocèle droite avec le diagnostic différentiel des Lymphangiome a été faite. Invesigations étaient essentiellement normal et le patient était prévue pour hydrocelectomy mais les résultats dans les opérations étaient différents.

RESULTATS: *A la chirurgie, il y avait une forte hypertrophie dur testicule droit avec cordon spermatique épaissi. Droit orchidectomie inguinale radicale avec l'excision du cordon spermatique droit et une adénopathie régionale a été réalisée. Histologie a révélé la LNH de grade intermédiaire nodulaire du testicule droit. Il a été traité avec une chimiothérapie systémique combinaison et a depuis été en rémission complète depuis plus de 10 ans.*

CONCLUSION: *lymphome testiculaire primaire est une tumeur rare et unique du testicule et est plus fréquente chez les hommes âgés de plus de 60 ans, mais devrait être considérée dans le diagnostic différentiel des tumeurs du testicule chez les groupes d'âge plus jeunes. Elle est curable au stade précoce avec multimodale de gestion approprié.* ***WAJM 2009; 28(6): 388–390.***

Mots-clés: *lymphome, du testicule, primaire, remise*

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Abbreviations: HIV, Human immunodeficiency virus; **NHL,** Non-hodgkin's lymphoma; **PLT,** Primary lymphoma of testis.

INTRODUCTION

Primary lymphoma of the testis is a rare tumour that accounts for about one percent of all Non Hodgkin's Lymphoma (NHL), about two percent of all extra nodal lymphomas and five percent of all testicular neoplasm.[1] Primary lymphoma of the testis is the most common testicular tumour in men between 60 and 80 years of age.[1] It has a high incidence of bilateral testicular involvement which is a unique feature and is now considered to be the most common bilateral neoplasm of the testes.[2] The prognosis is generally poor with progressive lymphomatous involvement.[3] It has a tendency for dissemination to non-contiguous extra nodal sites such as the Waldeyer's ring, skin and lungs. Central nervous system involvement has been reported in 10% of initial failure and 20% in subsequent relapses.[4] Thus lumbar puncture is warranted as one of the initial procedures at time of diagnosis.

We report the occurrence of this tumour rare in young people and with a very outcome.

Case Report

Patient was a Nigerian male civil servant who was 31 years old and single at presentation. His presenting complaint at the surgical outpatient department was a progressive painful right sided scrotal swelling of two years duration and progressive painful right groin swelling of one year. There was no history of weight loss, associated fever, night sweat, cough, vomiting, or nausea.

Examination revealed a young man with scrotal swelling and right hard, tender but mobile inguinal lymphadenopathy. The other system were essentially normal on physical examination.

A working diagnosis of right hydrocele with differential diagnosis of lymphangioma was made. Laboratory investigations revealed a normal haemogram and urinalysis. Human immunodeficiency virus (HIV) serology was negative, chest X-ray was normal. Patient was scheduled for hydrocelectomy but at surgery there was grossly enlarged hard right testis with thickened spermatic cord. Right radical inguinal orchidectomy with excision of the right spermatic cord and a regional lymph node was carried out.

Histology of excised organs and tissue revealed intermediate grade nodular NHL Based on the histology result, patient was referred to the Haematology department.

All test results were within normal limits. Abdominal ultrasound revealed no abnormality. Based on the histopathological diagnosis, clinical findings, results of abdominal ultrasound, and bone marrow aspiration, case was staged as 1EA testicular NHL using Ann Arbor classification.

The patient was treated with combination chemotherapy of Cyclophosphamide, along with allopurinol .The patient had six cycles of chemotherapy at 21days interval. Inguinal swelling gradually disappeared with systemic chemotherapy.

Six years after chemotherapy, patient came down with productive cough, night sweats, retrosternal pain with no history of chronic cough contact. Chest x-ray revealed left apical cavity while sputum smear for acid fast bacilli was positive.

He was treated with anti-tuberculosis drugs for eight months and he fully recovered. He complained of weak penile erection and premature ejaculation, was referred to the urology clinic where he was satisfactorily managed.

As at last visit he aged 42 years and married with two children. He is still in complete remission and attends follow up clinics regularly.

DISCUSSION

Testicular tumours are rare in Blacks while Nigeria has the lowest reported incidence at 0.1 per 100,000 per annum.[5–7] Non-germ cell tumours are reported to be more common in peoples of Negroid and Asian descent than in Caucasians while malignant lymphomas have been found to constitute 70% of non-germinal testicular tumours in Nigeria.[8]

Primary testicular lymphoma is uncommon, occurs principally in patients over 60 years old and is rare in men under 40 years; it is also unique in its incidence of bilateral involvement. However the case being reported presented at age 31years with unilateral right testicular lymphoma. Testicular lymphoma is commonly diffuse and aggressive[9] but this reported case is a nodular type which may have contributed positively to the good prognostic outcome. Findings in this patient are in keeping with a previous report that the most common mode of presentation of testicular cancers in Nigeria is scrotal swelling, usually accompanied by pain with a slight

Table 1: Laboratory Test Results Requested a are as shown:

Test	Result	Normal Range
Haematological Tests		
Haematocrit (%)	45	
Total white cell , /mm^3	3,700	
Platelet count ,/mm^3	157,000/mm^3	
Blood film	mild eosinophilia	
Bone marrow aspiration	essentially normal	
Serum Biochemistry		
Total Bilirubin , mg/dl	0.83	0–1
Direct Bilirubin, mg/dl	0.04	0–0.25
Alanine transaminase, u/L	17	0–40
Alkaline phosphatase, u/L	78	98–279
Fasting blood sugar, mg/dl	86	76–110
Uric acid , mg/dl	5.8	2.4–7
Cholesterol, mg/dl	144	50–200
Total Protein, g/dl	7.3	6.6–8.7
Albumin, g/dl	4.3	3.5–5

preponderance of right testicular involvement.[6]

There are no well documented aetiological or predisposing factors or any significant association existing between histories of trauma, chronic orchitis or cryptochidism and subsequent development of PLT.[3] It seems that the incidence of clinically apparent urogenital involvement by all lymphomas is approximately 5 to10%; it's much higher at autopsy, and NHL involves the urogenital tract significantly more often than Hodgkin's disease does.[10]

Secondary testicular involvement is not uncommon in advanced NHL cases either as part of terminal disease or in autopsy findings and up to 20% of patients dying due to disseminated NHL has been reported to have microscopic testicular invasion.[4, 11]

Non-Hodgkin's Lymphomas are classified by the Working Formulation of the United States National Cancer Institute as intermediate grade, diffuse large B-cell subtype, and high grade small non cleaved subtype in about 30% of the patients. There is no prognostic advantage for any pathological subtype.[2]

Malignant lymphoma of the testis can be confused with seminoma. Immunohistochemical stains have become the mainstay in differentiating carcinomas and lymphomas of the testis that cannot be distinguished by histology alone.[10] Magnetic resonance imaging is also a useful method of differentiating between testicular lymphoma and seminoma because it clearly shows tumour extension to the epididymis and spermatic cord.[12]

Scrotal and abdominal ultrasound is non-invasive, radiation free and indicated in clinically suggestive cases. Ultrasound is helpful in separating extra-testicular from intra-testicular lesions in painless scrotal swelling. It can also show whether a mass is cystic, solid or complex; features such as associated calcification, epididymal involvement, scrotal skin thickening and colour. Computerized tomography may be useful to rule out metastatic tumour.[13]

Serum lactic dehydrogenase levels have been correlated with tumour aggressiveness and should be measured because of both its values in prognosis and monitoring response to therapy. Other tumour markers such as serum human chorionic gonadotrophin and serum alpha fetoprotein are rarely elevated in PLT.

Needle aspiration or excision biopsy is necessary to establish a diagnosis. It is necessary to take samples from both testes because of the high incidence of bilateral involvement.

Treatment options for primary testicular lymphoma include orchidectomy, anthracycline based chemotherapy, prophylactic radiation therapy to contralateral testis and regional nodes as well as central nervous system prophylaxis with intrathecal chemotherapy.

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