**TESTICULAR ADNEXAL MESENCHYMAL TUMOURS IN IBADAN, NIGERIA: HISTOPATHOLOGIC SPECTRUM AND CHARACTERISTICS.**

\*Ogun GO1, Takure AO2, Ezenkwa US1, Chibuzor IN2.

1Department of Pathology, University of Ibadan/University College Hospital, Ibadan Nigeria

2Department of Surgery (Division of Urology), University of Ibadan/ University College Hospital, Ibadan

**\*Correspondence:**

Dr**.** G.O. Ogun,

Department of Pathology,

University of Ibadan/University College Hospital, Ibadan, Nigeria.

Email- olabiyiogun@gmail.com

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**ABSTRACT**

**Background:** Most testicular adnexal tumours are mesenchymal in origin and have a high tendency to be malignant. An earlier study from our centre found 10 cases, all malignant. The present study reviewed and updated testicular adnexal mesenchymal tumours diagnosed at the University College Hospital, Ibadan, Nigeria.

**Aims and Objectives:** To document the histopathologic spectrum and characteristics of testicular adnexal mesenchymal tumours diagnosed in our centre over a 20 year period and to compare these with earlier studies from our center and other parts of the world.

**Study Design:** This was a descriptive retrospective study.

**Setting of the study:** This study was carried out at the Departments of Pathology and Surgery (Urology Division), University of Ibadan/ University College Hospital, Ibadan Nigeria.

**Materials and Methods:** All testicular adnexal mesenchymal tumours from the files and records of the Department of Pathology, University College Hospital Ibadan from January 1, 1997 to December 31, 2016 were reviewed. Age, tumour size and histopathologic diagnosis were retrieved. The tumours were re-evaluated, immunohistochemistry was applied where appropriate and the tumours were reclassified histologically using the 2016 World Health Organisation classification of mesenchymal tumours of the spermatic cord and testicular adnexa. Descriptive statistics was applied on the data generated using SPSS version 20.

**Result:** Twenty-one cases of testicular adnexal mesenchymal tumours were found over the study period. Three (14.3%) were benign and comprised one case each of lipoma, leiomyoma, and fibrous pseudotumour. The remaining 18 (85.7%) cases were malignant. Rhabdomyosarcoma was the commonest tumour seen accounting for 12 (66.7%) of all the sarcomas followed by liposarcoma 3(16.7%). A case each of malignant peripheral nerve sheath tumour, fibrosarcoma and giant cell sarcoma were also documented. The overall mean age of the patients was 25.8±15.2 years with a range of 7-71 years, while the mean age of the patients with rhabdomyosarcoma was 22.0±8.7 years (8-42 years).

**Conclusion:** The outcome of this study suggests that the incidence of sarcomas of the testicular adnexa might be on the rise compared to previous report from our centre.

**Keywords:** Testicular adnexal, Mesenchymal tumour, Histopathology, Ibadan

**INTRODUCTION**

The majority of intrascrotal tumours are testicular in origin with few cases occurring in the spermatic cord and testicular adnexa1. About 50-65% of these spermatic cord and testicular adnexal tumours are of mesenchymal derivation2–4. It is often difficult to distinguish them from testicular tumours clinically.1 Population-based cancer registry data from an European study estimated the incidence of soft tissue sarcoma of the paratestis at about 0.03 per 100,000 population per year; this affirms the rarity of these tumours.5 Although the incidence of testicular and paratesticular tumours is higher in Caucasian populations when compared to Africans, adnexal tumours occur with higher percentage in Africans6. A study from Kano, Northern Nigeria found a total of 15 mesenchymal tumours out of 21 cases of paratesticular tumours seen over a 15-year period4. Previous studies from our centre, University College Hospital, Ibadan and Ile-Ife, both in Southwestern Nigeria showed that rhabdomyosarcoma was the commonest testicular adnexal sarcoma accounting for 7 out of 10 tumours seen over a 20-year and 17-year periods respectively in both centres6,7.

The aim of this study was to document the histopathologic characteristics of testicular adnexal mesenchymal tumours (tumour size and subtypes) diagnosed in our center and to compare these with an earlier study from our center and from other parts of the world.

**METHODS**

This was a descriptive retrospective study. All testicular adnexal mesenchymal tumours documented in the files and records of the Department of Pathology, University College Hospital, Ibadan from January 1, 1997 to December 31, 2016 were reviewed. Age, size of each tumour and histological diagnosis were reviewed. The cases were reviewed by (GOO and USE) independently and via a consensus where there was divergent opinion. The initial histological review was on some archival slides and then on newly sectioned haematoxylin and eosin stained slides. The tumours were subsequently classified using the 2016 World Health Organization(WHO) classification of mesenchymal tumours of the spermatic cord and testicular adnexa.8 Immunohistochemical (IHC) staining was applied on all the Rhabdomyosarcomas (Desmin) to demonstrate muscle differentiation in the tumour cells and on the Malignant Peripheral Nerve Sheath Tumour (S100) to show the neural origin of the tumour. The IHC staining helped to arrive at a final diagnosis. Descriptive statistics was performed on the data generated and presented as frequencies, mean and ranges using the Statistical Package for Social Sciences (SPSS) version 20.9 This study was conducted in compliance with the guidelines of the Helsinki declaration on biomedical research in human subjects. Confidentiality of the identity of the patients and personal health information was maintained as no name identifier was used in retrieving the cases from the records.

**RESULTS**

Twenty-one cases of testicular adnexal mesenchymal tumours were identified over the study period. **Table 1** shows the age distribution of the study population. The mean age of the patients was 25.8±15.2 years (range 7-71 years). Overall, the tumour sizes ranged from 3cm to 17cm in the widest dimension with a median size of 12.9cm.

Three (14.3%) of the tumours were benign while 18(85.7%) were malignant. The benign tumours consisted of one case each of lipoma, leiomyoma and fibrous pseudotumour. Rhabdomyosarcoma was the commonest malignant tumour, accounting for 12 (66.7%) of all the sarcomas followed by liposarcoma 3(16.7%); there was one case each of fibrosarcoma, giant cell sarcoma and malignant peripheral nerve sheath tumour **(Table 2)**. The mean age of the patients with rhabdomyosarcoma was 22.0±8.7 years (range 8-42 years), with eight (66.7%) of them being 25 years old or younger. Three variants of rhabdomyosarcoma were identified – pleomorphic rhabdomyosarcoma (3 cases); embryonal rhabdomyosarcoma (3 cases); and alveolar rhabdomyosarcoma (1 case). The remaining 5 cases of rhabdomyosarcoma could not be further characterized. One of the cases of embryonal rhabdomyosarcomas showed testicular invasion on histology. Two out of 3 of the liposarcomas observed occurred in adolescents of 18 and 20 years of age while the third case occurred in a 55 year old man. A case of malignant peripheral nerve sheath tumour (MPNST) was seen in a 17 year old who had no family history or clinical finding suggestive of neurofibromatosis. **Table 3** shows comparison of various tumour types across different studies. **Figure 1** shows gross histopathology of a typical adnexal tumour in a patient who had orchidectomy for a scrotal mass diagnosed clinically but proved to be benign on subsequent histopathological evaluation.

**DISCUSSION**

The rarity of testicular adnexal mesenchymal tumours has been consistently attested to by the very few cases reported from various studies2–4,6,7. Our result of 21 cases of these tumours over a 20-year period in this study as against 10 cases previously reported in our center over a similar time interval suggests that these conditions are now diagnosed more frequently.6  This increased frequency has also been demonstrated in a comprehensive European Study by Trama et al10. The explanation for this trend cannot be easily deduced. We suggest that it may be related to the increasing population and improved health-seeking behavior of patients. In this current study we have shown that these tumours affect all age groups but most especially children and adolescents. This finding is similar to that by Alhaji *et al* in Kano, Northern Nigeria.4 In that study 12(75%) of the tumours occurred in the 0-9 and 10-19 age groups, while the remaining three cases occurred in the 40-49 and 50- 59 year age groups4. In contrast, 8 cases (80%) of the paratesticular tumours documented by Salako *et al* occurred among individuals aged 15 – 44 years while 2 (20%) occurred in the age group 45 – 64 years.7

The finding of more sarcomas rather than benign tumours in this study is in contrast with studies by Lioe and Biggart,2 Birmingham et al,3 and Alhaji et al4 but similar to that by Salako et al7(See **Table 3**). No benign tumours were documented in the previous study from our center.6 Different studies have shown differences in which subtype of sarcoma is commonest. In a review by Khoubehi *et al*, leiomyosarcoma was the commonest (32%) followed by rhabdomyosarcoma (24%)1, while liposarcoma was reported as being commonest by Gigantino *et al*, constituting 30% of all sarcomas reviewed, however, that study considered cases occurring in older men with average age of 55 years11 Our data shows that rhabdomyosarcoma is the commonest sarcoma of the testicular adnexal followed by liposarcoma and this is in agreement with the earlier study from our center and another local study from Ile-Ife, both in Southwest, Nigeria.6,7

Testicular adnexal rhabdomyosarcoma occurs more frequently in children and adolescents while liposarcomas are more likely to be seen in the older age groups.12–14 In the present study, all the rhabdomyosarcomas and 2 out of 3 cases of liposarcomas were seen in young adults Furthermore, cases of rhabdomyosarcoma occurring in the elderly have been documented.15 Liposarcomas constitute the commonest soft tissue sarcoma seen in man with about 3-7% of these being observed in the testicular adnexal11; they are believed to originate from the spermatic cord as part of the retroperitoneal fat that accompanies the cord during the descent of the testis16. Pathologically, it could be difficult to ascertain the origin of paratesticular liposarcomas, because they attain enormous size as at the time of diagnosis16. The tendency of large size tumour in this location is illustrated by the average size of tumours in this study being 12.9cm.

Other sarcomas found in this study include malignant peripheral nerve sheath tumour (MPNST) and giant cell sarcoma which are quite rare in the literature8. Few cases of neurofibroma in this site have been documented4,17, and their malignant variant is even more uncommon8. The age of the index patient in this study with MPNST is similar to that documented in the literature of a 15 year old boy with MPNST who had associated paratesticular ganglioneuroma18. Giant cell sarcoma of soft tissue is remarkably rare. We did not find any previous report of giant cell sarcoma of the testicular adnexa.

Though, from literature lipoma is the commonest benign mesenchymal tumour of the testicular adnexa, we found only one case in this study. 2,8,16 Furthermore, we found one case each of Leiomyoma and fibrous pseudotumour, these tumours have been reported previously at this site in another study3.

Patients may benefit from intra-operative pathologic consultation to determine whether the tumour is benign or malignant, which will prevent radical surgery such as orchidectomy that may prove unwarranted following examination of the formalin fixed paraffin embedded cut sections.19 This is illustrated in Figure 2 in a patient who had orchidectomy for a scrotal mass and which ultimately turned out to be a benign tumour.

Possible limitation of this study includes its hospital-based nature. As such, not all cases of these tumours might have been documented especially the benign tumours. Despite this, the data presented here is similar to studies from other populations though with some minor differences.

In conclusion, testicular adnexal mesenchymal tumours appear to be currently more frequently diagnosed at our centre, most of them being sarcomas. Rhabdomyosarcoma remains the commonest sarcoma seen and occurs more commonly in children and young adults. Effort to recognize the benign cases is advocated to avoid over-treatment of the patients with unnecessary orchidectomy.

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**Table 1:** Age distribution of the study population

|  |  |  |
| --- | --- | --- |
| Age Range (years) | Frequency | Percentage (%) |
| 0-9 | 1 | 4.8 |
| 10-19 | 7 | 33.3 |
| 20-29 | 7 | 33.3 |
| 30-39 | 2 | 9.5 |
| 40-49 | 2 | 9.5 |
| ≥50 | 2 | 9.5 |
| Total | 21 | 100 |

**Table 2:** Histopathological classification of tumour subtypes

|  |  |  |
| --- | --- | --- |
| **Tumour subtype** | **Frequency** | **Percentage** |
| Rhabdomyosarcoma | 12 | 57.1 |
| Liposarcoma | 3 | 14.3 |
| Malignant peripheral nerve sheath tumour | 1 | 4.8 |
| Fibrosarcoma | 1 | 4.8 |
| Giant cell sarcoma | 1 | 4.8 |
| Lipoma | 1 | 4.8 |
| Leiomyoma | 1 | 4.8 |
| Fibrous pseudotumour | 1 | 4.8 |
| **Total** | 21 | 100 |

**Table 3**: Spectrum of mesenchymal testicular adnexal tumours from different studies.

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| Tumour Type | Junaid, 1982  (Ibadan, Nigeria)6 | Lioe & Biggart 1993 (Northern Ireland)2 | Birmingham et al, 2012 (Spain)3 | Alhaji et al, 2016 (Kano, Nigeria)4 | Salako et al  2010  (Ile-Ife, Nigeria)7 | Present study (Ibadan, Nigeria) |
| Lipoma | - | 27 | - | 1 | - | 1 |
| Leiomyoma | - | 6 | 5 | 3 | 1 | 1 |
| Fibroma | - | 1 | 1 | - | - | 1 |
| Haemangioma | - | 2 | - | - | - | - |
| Neurofibroma | - | 1 | - | 4 | - | - |
| Rhabdomyosarcoma | 7 | 2 | - | 3 | 7 | 12 |
| Liposarcoma | - | 2 | 3 | 1 | 1 | 3 |
| Leiomyosarcoma | 1 | 2 | - | 2 | 1 | - |
| Fibrosarcoma | 1 | 1 | - | 1 | - | 1 |
| MFH | - | 1 | 1 | - | - | - |
| MPNST | - | - | - | - | - | 1 |
| Giant cell sarcoma | - | - | - | - | - | 1 |
| Undifferentiated Embryonic sarcoma | 1 | - | - | - | - | - |
| **Total** | **10** | **45** | **10** | **15** | **10** | **21** |
|  |  |  |  |  |  |  |

MFH: Malignant fibrohistiocytoma; MPNST: Malignant peripheral nerve sheath tumour

**Figure 1**- Gross morphology of the cut section of a typical adnexal tumour in a patient who had orchidectomy for a scrotal mass diagnosed clinically.



**Normal testicular tissue**

**Paratesticular tumour**

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