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Case Series

Urology Section

Rare Non-germ Cell Testicular Tumors: A Series of Three Cases

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ABSTRACT

Testicular neoplasms are the most common malignancy among men aged 20-40 years. More than 95% are germ cell tumours and 5% are sex cord stromal tumours. The most common presentation is a painless scrotal lump. Sometimes, it may present as a painful lump, mimicking an inflammatory lesion. Case 1, a 34-year-old male presented with right testicular pain and a lump for one month, with normal tumour markers. Clinically, it appeared as a testicular malignancy, but with normal tumour markers, it mimicked an inflammatory lesion, creating a management dilemma. A Computed Tomography (CT) scan ruled out retroperitoneal lymph node metastasis. Right high inguinal orchidectomy with frozen section was performed, suggesting malignancy. Immunohistochemistry confirmed leiomyosarcoma. Case 2, an 82-year-old male presented with a painless right testicular lump for one month, with a history of acute urinary retention secondary to benign prostatic enlargement {treated with Transurethral Resection of the Prostate (TURP)}. Clinically, it was a hard lump suggestive of malignancy, confirmed on ultrasound, although tumour markers were normal. A CT scan was negative for lymphadenopathy. Right high inguinal orchidectomy was performed. Histopathological examination showed non-Hodgkin's lymphoma; the patient subsequently underwent chemotherapy. Case 3, a 29-year-old male complained of right testicular pain. Examination revealed a tender nodule (1×1 cm) on the posterolateral aspect of the right testis, clinically appearing as a firm mass, confirmed on ultrasonography, although tumour markers were misleadingly normal. High inguinal orchidectomy was performed, with histopathological examination positive for intratesticular adenomatoid tumour. High inguinal orchidectomy was the treatment of choice in all three cases, followed by adjuvant chemotherapy or radiotherapy depending on histopathology. Conclusion: Testicular tumours may mimic inflammatory lesions, creating management dilemmas. Thorough clinical examination and investigations are required for planning and management. Normal serum tumour markers and ultrasound cannot rule out malignancy.

Keywords: Adenomatoid, Leiomyosarcoma, Non germ cell tumour, Non-hodgkins lymphoma

INTRODUCTION

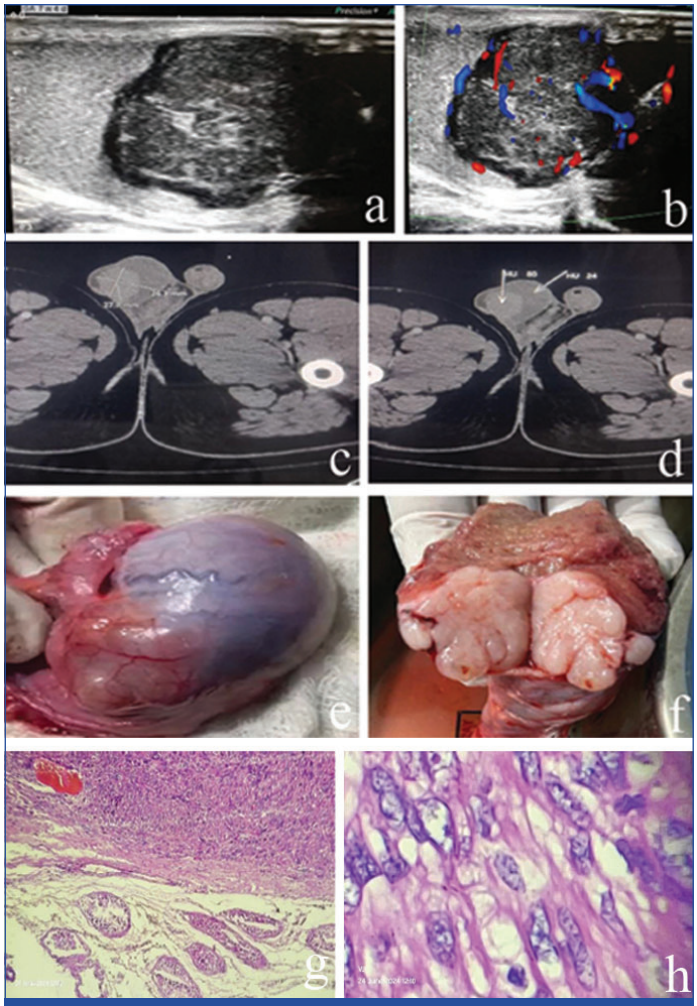
Testicular neoplasms are the most common malignancy among men aged 20-40 years. The aetiology is multifactorial, encompassing various genetic and environmental factors. Testicular tumours account for 1% of male cancers and 5% of urological malignancies [1]. Testicular tumours are classified by cell type. More than 95% are germ cell tumours and 5% are sex cord stromal tumours. Germ cell tumours are further classified into seminomatous and non seminomatous tumours [1]. Sex cord tumours are generally benign, but 5% are malignant. Genitourinary sarcomas are very common sarcomas in adults, with liposarcoma followed by leiomyosarcoma being the most common histological subtypes. The most common sites are the spermatic cord and epididymis, although primary mesenchymal tumours of the testes are very rare [2]. Intratesticular adenomatoid tumours are rare, generally arising from paratesticular structures such as the epididymis. Diagnosis relies entirely on immunohistochemistry and pathological assessment [3]. Early detection through thorough clinical examination and radiological investigation provides a 90-95% cure rate. Testicular tumours are among the most curable cancers. The present case series discusses three very rare testicular tumours that clinically mimicked inflammatory lesions and had normal tumour markers.

Case 1

A 36-year-old male presented with one month of right testicular pain, gradual in onset, dull aching and not associated with vomiting or fever. There was no history of trauma, surgery, or family history of malignancy. Examination revealed a normal left testis and a 2×1 cm lump on the posterior aspect of the right testis, with mild tenderness. Ultrasound of the inguinoscrotal region showed a heterogeneous intratesticular mass (25×25 mm) in the right testis with high vascularity. Tumour markers {Lactate Dehydrogenase (LDH), β-Human Chorionic Gonadotropin (β-HCG), α-Fetoprotein (AFP)} were within normal limits. Ultrasound showed a heterogeneous intratesticular mass (25×25 mm) in the right testis with high vascularity [Table/Fig-1a,b]. CT scan of the abdomen and pelvis showed a 27.6×20.6 mm mass in the right testis (45 HU) [Table/Fig-1c,d]. The left testis was unremarkable, with no abdominal, pulmonary, or pelvic node enlargement. These findings suggested a malignant mass, leading to a planned high inguinal orchidectomy. Intraoperatively, frozen section from the mass was consistent with malignant sarcoma. Gross examination showed a soft, lobulated mass arising from the upper pole of the testis [Table/Fig-1e], with a pale-tan, firm mass on cut section, without haemorrhage or necrosis [Table/Fig-1f]. Histopathological examination showed spindle-shaped cells with cigar-shaped nuclei and eosinophilic cytoplasm, with areas of necrosis and mild atypia, suggesting intratesticular sarcoma. Immunohistochemistry (IHC) ruled out other differentials (liposarcoma, leiomyoma, mesothelioma, rhabdomyosarcoma) [Table/Fig-1g,h], being positive for desmin [Table/Fig-2a], caldesmon [Table/Fig-2b] and Smooth Muscle Actin (SMA) [Table/Fig-2c] and negative for CD34 [Table/Fig-2d], cytokeratin [Table/Fig-2e] and myogenin [Table/Fig-2f], consistent with leiomyosarcoma. Postoperative recovery was uneventful and the patient was discharged on postoperative day 2. He is currently under follow-up.

Case 2

An 82-year-old male presented with a two-month history of right testicular swelling, gradual in onset and painless, not associated with fever, vomiting, or weight loss. He had a history of acute urinary retention secondary to benign prostatic hyperplasia, treated with TURP. There was no history of trauma or family history of testicular tumours. Examination revealed a normal left testis and an enlarged,



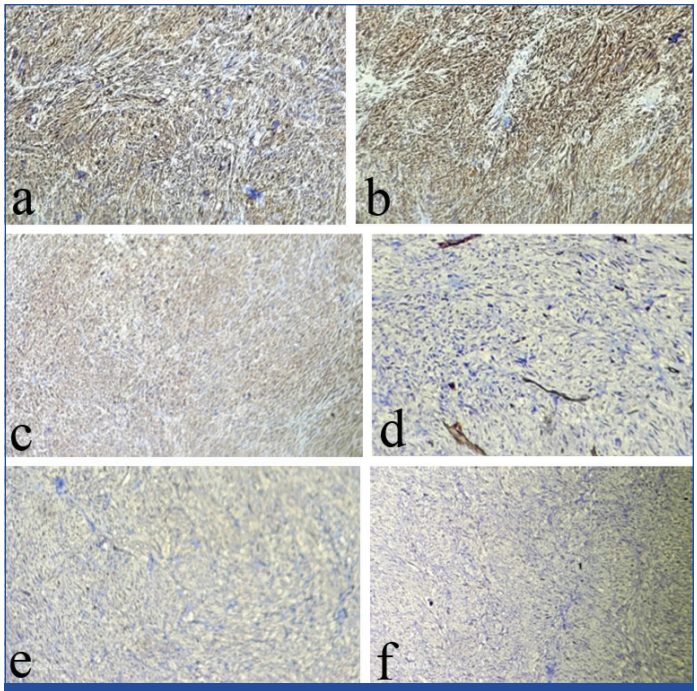
[Table/Fig-1]: a,b) USG B/L inguinoscrotal region suggestive of a heterogeneous intratesticular mass of size 25*25 mm in Rt testis with high vascularity; c) CT abdomen pelvis done which showed a mass of size 27.6*20.6 mm; d) Rt testis with 45HU; e) Gross examination of specimen suggestive of soft lobulated mass arising from the upper pole of the testis; f) On cut section it shows a pale-tan firm mass without areas of haemorrhage and necrosis; g) Spindle shape cells with cigar shaped nuclei and eosinic cytoplasm (H&E, 10x); h) Spindle shape cells and cigar-shaped nuclei (H&E, 40x).

firm-to-hard right testis. No lymphadenopathy was noted. Tumour markers (LDH, β-HCG, AFP) were within normal limits. Ultrasound of the scrotum showed an enlarged right testis with heterogeneous echogenic deposits, with maintained vascularity, suggestive of lymphoma or testicular tumour. CT scan of the abdomen and pelvis showed no enlarged retroperitoneal or abdominal lymph nodes. Chest X-ray was negative for metastasis. Positron Emission Tomography (PET) scan showed no Fluorodeoxyglucose (FDG) avid distant metastatic lesions. High inguinal orchidectomy was performed under spinal anaesthesia and the specimen was sent for histopathological examination.

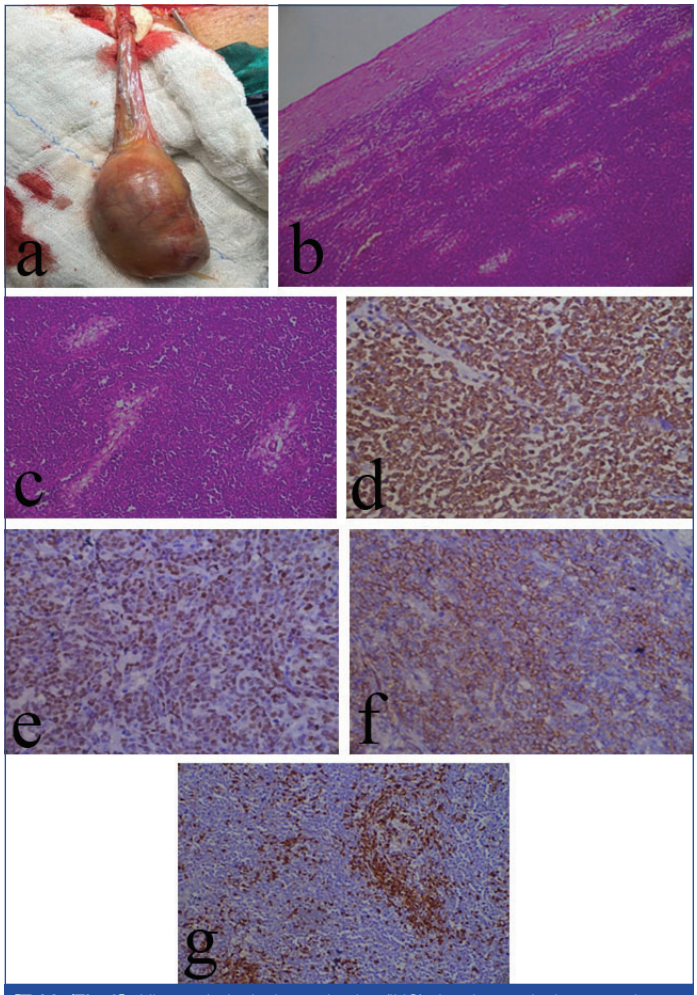
The IHC staining was performed to rule out other possibility of seminoma, embryonal carcinoma, myeloid neoplasm. IHC was positive for B-cell Lymphoma 2 (BCL2) [Table/Fig-3a], BCL6 [Table/Fig-3b], Cluster of Differentiation (CD)-20 [Table/Fig-3c], CD-3 Reactive T cell [Table/Fig-3d] suggestive of poorly-differentiated non-hodgkins lymphoma. The patient is undergoing chemotherapy with a Rituximab, Cyclophosphamide, Hydroxydaunorubicin (Doxorubicin), Oncovin (Vincristine) and Prednisone (R-CHOP) regimen.

Case 3

Description: A 29-year-old male presented with right testicular pain, gradual in onset, not associated with fever, nausea, vomiting, or weight loss. There was no history of trauma or family history. Examination showed a normal left testis and a 1×1 cm mass on the posterolateral aspect of the right testis, with tenderness suggestive of an inflammatory lesion. Ultrasound of the inguinoscrotal region

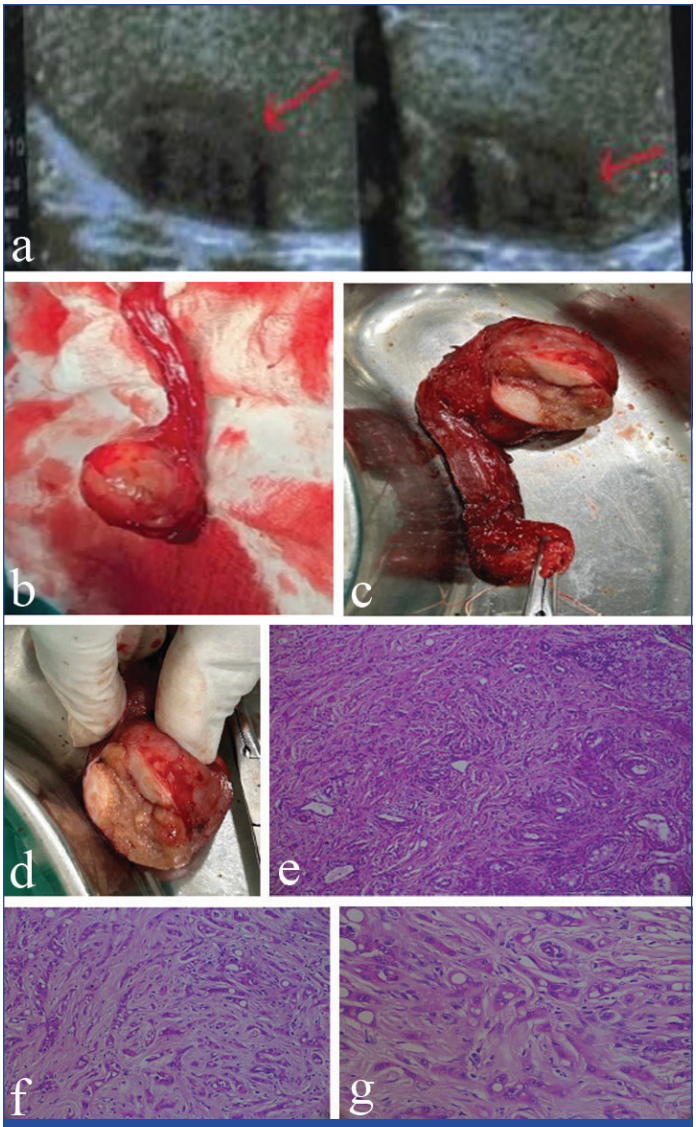


[Table/Fig-2]: a) Desmin positive under 10x; b) Caldesmonpositive under 10x; c) SMA100 Positive under 10x; d) CD34 Negative under 10x; e) Cytokeratin negative-under 10x; f) Myogenin negative under 10x.



[Table/Fig-3]: Histopathological examination (IHC) showing testicular parenchyma replaced by sheets of diffuse large lymphoid cells, suggestive of poorly differentiated Non-Hodgkins lymphoma. a) Gross specimen; b) Tumour cell in low power 10x showing diffuse sheets of lymphoid cells; c) Tumour cells in high power 40x showing diffuse sheets of lymphoid cells; d) BCL2 Positive under 400x; e) BCL6 Positive under 400x; f) CD20 Positive under 400x; g) CD3 Reactive T cell under 400x.

showed a normal left testis (4.1×3.2×1.8 cm) and a heterogeneous mass (16×13×8 mm) in the right testis (4.3×3×2.2 cm) [Table/Fig-4a]. Tumour markers (LDH, β-HCG, AFP) were within normal limits. Contrast-Enhanced Computed Tomography (CECT) of the abdomen



[Table/Fig-4]: a) USG Inguinoscrotal showing 16×13×8 mm heterogeneously enhancing lesion in right testis marked with red arrow; b) Gross specimen of left testis; c) Cut-section of tumour showing tan gritty mass; d) Hard gritty mass on cut-section; e) Tumour cells on low magnification 10x showing numerous tubulo-glandular structure lined by cuboidal cells and testicular parenchyma with areas of fibrosis. Tumour composed of numerous tubule-glandular structure lined by cuboidal cells having vesicular nuclei and prominent nucleoli; f) Tumour cells on high power 40x showing tumour cells with vesicular nuclei and abundant cytoplasm; g) Tumour cells in 40x showing vesicular nuclei and prominent nucleoli.

and pelvis showed no enlarged abdominal or retroperitoneal lymph nodes. Chest X-ray was negative for metastasis. PET scan showed no FDG-avid distant metastatic lesions. High inguinal orchidectomy was performed. The patient is currently under follow-up with no clinical or radiological evidence of contralateral testicular lesions. ([Table/Fig-4b] shows the gross specimen; [Table/Fig-4c] and [Table/Fig-4d] show the whitish, hard and gritty mass on cut section).

The HPE showing testicular parenchyma with areas of fibrosis with tumour composed of numerous tubuloglandular structure lined by cuboidal cells. These have vesicular nuclei, prominent nucleoli and abundant cytoplasm suggestive of adenomatoid tumour.

DISCUSSION

Testicular tumours are most common in men aged 15-45 years. The majority (95%) are germ cell tumours, with 5% being sex cord tumours. The most common presentation is a painless scrotal lump. In the present case series, all three patients presented with both pain and a lump. Case 1 presented with acute urinary retention and right testicular pain (treated with TURP), later evaluated for a testicular mass. Histopathological examination and IHC were consistent with intratesticular leiomyosarcoma. While paratesticular leiomyosarcomas are common, intratesticular leiomyosarcomas are rare. The most common age of presentation is the fourth to seventh decades [4]. The aetiology is unclear, often associated with radiation therapy or anabolic steroid use, neither of which were present in the present case. Tumours may spread via local invasion, lymphatic dissemination, or haematogenous metastasis [5]. A similar case was published by Bakhshi GD et al., [6], presenting in the sixth decade with high LDH, unlike this case, which presented in the third decade with normal markers. In all reported cases of testicular leiomyosarcoma, high inguinal orchidectomy is the treatment of choice, with few cases receiving adjuvant chemotherapy or Retroperitoneal Lymph Node Dissection (RPLND) [Table/Fig-5] [6-11]. In Case 2, the patient presented with a painless right testicular mass, without vomiting, fever, weight loss, or lymphadenopathy. High inguinal orchidectomy was performed, with histopathological examination positive for poorly differentiated non-Hodgkin's lymphoma {Testicular Diffuse Large B-Cell Lymphoma (T-DLBCL)}. A literature review by Medina A et al., [12] included 15 cases of testicular non-Hodgkin's lymphoma, with a median age of 69 years and most commonly presenting as testicular swelling. CNS involvement occurred in 6 cases, with DLBCL being the most common variant (80%). The contralateral testis relapse rate was 13.3% [12]. The present case presented with a similar age and symptoms. T-DLBCL accounts for 1-2% of non-Hodgkin's lymphomas, with an incidence of 0.09–0.26/100,000 population, representing about 5% of all testicular malignancies. T-DLBCL arises primarily in the immune-privileged site of the testis, where the blood–testis barrier protects the testicular tissue [13]. The standard of care is orchiectomy followed by immunochemotherapy with six cycles of R-CHOP or a similar regimen. The patient is currently receiving chemotherapy and is under regular follow-up. In Case 3, the patient presented with left testicular pain, mimicking an inflammatory lesion. High inguinal orchidectomy was performed, with histopathological examination revealing an intratesticular adenomatoid tumour. Adenomatoid tumours are benign, mostly arising from paratesticular structures [14], but intratesticular adenomatoid tumours are rare, creating diagnostic dilemmas due to their atypical location. A similar case was presented by Al Diffalha S et al., [15], undergoing high inguinal orchidectomy, the treatment of choice as most intratesticular tumours are malignant. European Association of Urology (EAU) guidelines suggest that testicular organ-sparing surgery is possible in special cases, such as patients with a tumour in a solitary testis, bilateral testicular cancers, or metachronous contralateral tumours [16]. In all three cases, inguinoscrotal ultrasound was performed as the primary investigation. CECT of the abdomen and pelvis ruled out intra-abdominal or retroperitoneal metastasis and lymphadenopathy. Chest X-ray ruled out lung metastasis. Tumour markers were negative in all three cases. The standard treatment for testicular tumours is high inguinal orchidectomy, performed in all three cases, followed by chemotherapy or radiotherapy depending on the histological type.

Author names	Age	Stage	Treatment	Outcome
Bakhshi GD et al., [6]	30 y	1	HIO	Survived
Yachia D et al., [7]	55 y	1	HIO	Survived
Sattary M et al., [8]	27 y	1	HIO	Survived
Wakhlu A and Chaudhary A [9]	8 m	1	HIO +CTx	Survived
Froenchner M et al., [10]	32 y	1	HIO+RPLND	Survived
Canales BK et al., [11]	60 y	1	HIO+RT	Survived
Present case	36 y	1	HIO	Survived

[Table/Fig-5]: Comparison of similar studies with present study [6-11].
HIO: High inguinal orchidectomy; CTx: Chemotherapy; RPLND: Retroperitoneal lymph node dissection; RT: Radiotherapy

CONCLUSION(S)

Testicular tumours may mimic inflammatory lesions, creating treatment and management dilemmas, especially when arising from unusual locations. Thorough clinical examination, radiological investigations and blood tests are necessary for management

planning. Normal serum tumour markers cannot rule out malignancy.

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