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CASE REPORT

Paratesticular Liposarcoma of the Spermatic Cord: A Case Report and a Review of the Literature

Liposarcome paratesticulaire du cordon spermatique: Un rapport de cas et une revue de la littérature

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ABSTRACT

BACKGROUND: **Paratesticular liposarcomas are uncommon and in view of this most practitioners would be unfamiliar with the diagnosis and management.**

OBJECTIVE: **To report a case of paratesticular liposarcoma, a rare tumour and to review the literature on paratesticular liposarcoma.**

METHODS AND RESULTS: **A 77-years old man with paratesticular liposarcoma originating from the left spermatic cord is reported. This patient noticed the lump eight years prior to its excision. The tumour was originally thought to be benign lipoma and was left alone and its management was based upon the principle of watchful waiting. The tumour was excised as an additional procedure when the patient underwent trans urethral resection of prostate following lower urinary tract symptoms and retention of urine and at this stage the 'lipomatous' lump was noticed to have grown bigger over recent months. The patient underwent a successful surgical excision (a radical excision of the testis and surrounding mass) and has remained under surveillance by the Regional Oncologists. Literature review revealed that 109 cases of para-testicular liposarcoma had previously been reported and radical excision with a wide resection margin is the recommended surgical approach to its management.**

CONCLUSION: **Radical surgical excision with wide resection margins is the most appropriate approach to the management of para-testicular liposarcoma and the patients should be referred to the Oncologist. WAJM 2011; 30(6): 447–452.**

Keywords: **Paratesticular liposarcomas, Spermatic Cord, Trojani grading, well differentiated, sclerosing.**

RÉSUMÉ

CONTEXTE: Liposarcomes paratesticulaires sont rares et en vue de cette la plupart des praticiens seraient pas familiers avec le diagnostic et la gestion.

OBJECTIF: Rapporter un cas de liposarcome paratesticulaire, une tumeur rare et revue de la littérature sur le liposarcome paratesticulaire.

MÉTHODES ET RÉSULTATS: Un homme de 77 ans avec un liposarcome paratesticulaire provenant du cordon spermatique gauche est rapporté. Ce patient a remarqué les huit forfaitaires ans avant son excision. La tumeur a été initialement pensé pour être un lipome bénin et a été laissé seul et de sa gestion a été fondée sur le principe de l'attente vigilante. La tumeur a été excisée comme une procédure supplémentaire lorsque le patient a subi une résection trans urétrale de la prostate à la suite des symptômes du bas appareil urinaire et de rétention d'urine et, à ce stade de la «lipomateuse 'forfaitaire a été remarqué avoir grossi ces derniers mois. Le patient a subi une excision chirurgicale réussie (une excision radicale du testicule et de la masse environnante) et est resté sous surveillance par les oncologues régionaux. Revue de la littérature a révélé que 109 cas de para-testiculaire liposarcome avait déjà été signalé et l'excision radicale avec une marge de résection large est l'approche recommandée chirurgicale à sa gestion.

CONCLUSION: Radical excision chirurgicale avec des marges de résection larges est l'approche la plus appropriée à la gestion de la para-testiculaire liposarcome et les patients doivent être référés à l'oncologue. **WAJM 2011; 30 (6): 447–452.**

Mots-clés: Paratesticulaire liposarcomes, cordon spermatique, le classement Trojani, bien différenciés, sclerosin.

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Abbreviations: DDL, Dedifferentiated lipo-sarcomas **LMS, WDL,** Well differentiated liposarcomas; **WDLPS**

INTRODUCTION

Paratesticular liposarcomas are uncommon and in view of this most practitioners would be unfamiliar with the diagnosis and management. A case of para-testicular liposarcoma arising from the spermatic cord is reported with a review of the literature.

Case Report

A 77-year old man had transurethral resection of prostate as a result of lower urinary tract symptoms and retention of urine. The patient also had a mass in his left hemi-scrotum which was clinically considered to be multiple lipomata of the cord. He noticed the lump in his left hemiscrotum eight years prior to his operation. He was seen in the outpatient clinic one and half years prior to the operation. At that time the lump on examination was considered to be intrascrotal lipoma of the cord and in view of the fact that the lump had remained the same size for many years it was felt that the lump should be left alone. However, at the time of the prostatic resection the patient said he had noticed a recent increase in size of the lump. In view of this it was decided to excise the lump which was provisionally considered to be a lipoma. During the excision, the mass was thought to be a complex multi-loculated lipoma which surrounded the cord and it was felt that the best way to ensure a complete excision was to excise the mass together with the left testicle in order to avoid leaving behind residual lipomatous tissue due to the fact that separation of the multilobulated fatty lump from the cord was difficult. The left testis, the fatty lump and the cord were excised together. During the dissection the mass was easily shelled out from the subcutaneous tissue because it was not attached to it. There was no connection of the mass to the right hemiscrotal sac or contents of the right hemiscrotum. The right testis, epididymis and cord felt normal. The mass which was obviously confined to the left hemiscrotum was also noted to be related to the cord but palpably not attached to the left testis but encasing the spermatic cord. The post operative recovery was unremarkable and the patient was discharged home.

On macroscopic examination, the left testis with the attached tumour together weighed 443 grams and measured 19.0 x 12.0 x 6.0 centimetres. The attached testis measured 4.0 x 3.0 x 2.5 centimetres (See Figure 1). The surface of the tumour was nodular and partly encapsulated. The cut surfaces were solid, lobulated and yellow. With serial slicing it was observed to have variable consistency and one of the nodules showed a small area of haemorrhage and myxoid change. Microscopically, the scrotal tumour mass, revealed features of lipoma-like morphology in most places. However, focally sclerosing areas were identified. There were areas of necrosis. There were frequent lipoblasts. Immuno-histological staining for S-100 was positive in lipoblasts. (See Figures 2 to 6 which illustrates various aspects of the microscopic features). The appearances were those of a well differentiated sclerosing liposarcoma/(modified Trojani/ FNCLCC grade 1 neoplasm). Histology of the prostate was consistent with benign prostatic hyperplasia.

A post operative CT scan of the chest, abdomen and pelvis was performed two weeks later but this did not show any evidence of a metastatic lesion.

The patient remained well without any evidence of recurrence and was also voiding well six months after his operation. He has been referred to the Regional Oncology Centre and he would be jointly followed up by our Urology team and the Oncology Team.

DISCUSSION

Paratesticular liposarcomas are rare tumours. These tumours are typically reported as isolated cases or as components of larger studies of liposarcomas and hence literature on the biological behaviour of this rare tumour has been anecdotal. To our knowledge this is the 110th case of liposarcoma of the spermatic cord to be reported in the literature. .

Massoud and associates[1] reported the 101st case of liposarcoma of the spermatic cord in December 2005, since then 8 new cases have been reported.[2–7]

Montgomery and Fisher[8] reported on 30 cases of paratesticular liposarcomas in men aged 41–87 years (mean 63 years; median 65 years) that involved the spermatic cord (23, 76%), testicular tunics (6, 20%), and epididymis (1, 4%). The tumours ranged from 3 to 30cm (mean 11.7 cm; median 10 cm). Nineteen were well differentiated liposarcomas (WDLs) and 10 were dedifferentiated liposarcomas (DDLs); – five with high-grade and five with low-grade dedifferentiation. One was a myxoid/round cell liposarcoma with 70% round cell areas. All patients were treated by radical orchiectomy. One patient with WDL received radiation after his second recurrence and the myxoid/ round cell liposarcoma received radiation and chemotherapy. Follow-up information was available for 16 of the patients, including 10 WDLs (range 24–216 months, mean 106 months), 5 DDLs (14–30 months, median 24 months), and for the myxoid/round cell liposarcoma (14 months) range for all cases 14 months to 22 years; mean 87 months, median 36 months). Six of the WDLs (60%) recurred at 2, 4, 6, 10, 18, and 21 years (median 8 years). The lesion which recurred at 18 years displayed foci of high-grade dedifferentiation in the recurrence even though the patient was disease free at 19 years. One patient with WDL developed two recurrences at 4 and 7 years, and another had six recurrences over a period of 17 years. Only one example of DDL recurred, at 30 months, another patient, who refused therapy for 15 years, had a primary tumour 30 cm in diameter, displayed pulmonary metastases 1 month after excision, and died after 14 months. The patient with MRCL had abdominal metastases after 1 year and was alive at 14 months. Montgomery and Fisher[8] concluded that WDL had a prolonged course with recurrences in more than half the cases, sometimes late. There were no metastases and the overall prognosis was good. One DDL recurred and only one of five (20%) developed metastases, but the mean follow-up for DDL was only 24 months.

Folpe and Weiss observed that liposarcomas may have low grade leiomyosarcomatous differentiation, which does not affect prognosis.[9] Folpe and Weiss[9] reported their experience with nine cases, (seven male, two female) with age ranges between 42 years and 65 years (mean 54 years). The tumours were

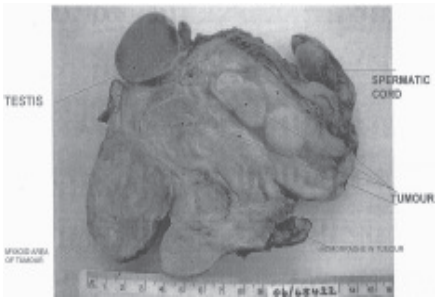

Fig. 1: A photograph of the cut section of the specimen showing the macroscopic appearance of the tests, large multi-loculated tumour and the spermatic cord.

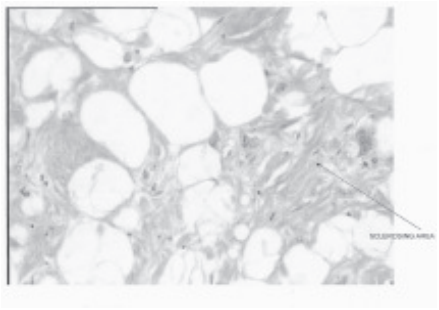

Fig. 2: Microphotograph showing sclerosing areas in the tumour and increased cellularity. Haematoxylin and Eosin stain. Magnification 100x.

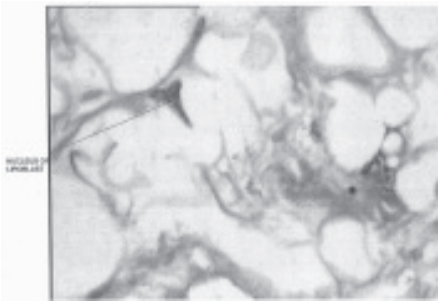

Fig. 3: Microphotograph of liposarcoma showing lipoblasts. Magnification 200x.

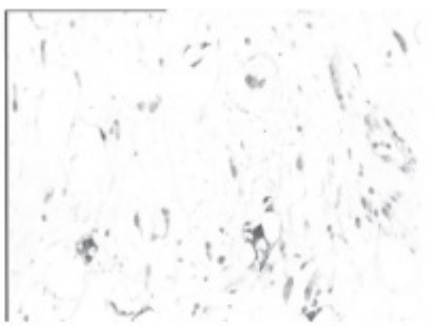

Fig. 4: Microphotograph showing immunohistological staining for S–100 positive in lipoblast Haematoxyling and Eosin staining. Magnification 200x.

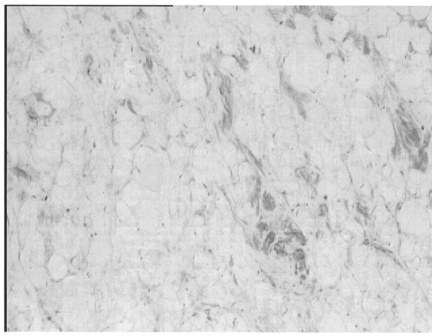

Fig. 5: Microphotograph showing lipoid areas. Haematoxylin and Eosin stain. Magnification 25x.

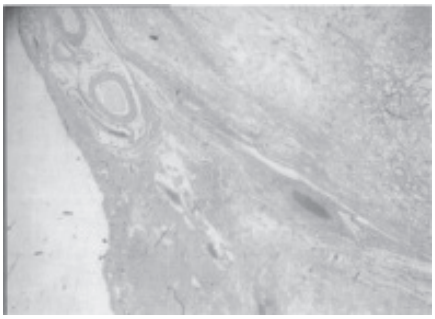

Fig. 6: Microphotograph showing tumour and adjacent spermatic cord tissue. Haematoxylin and Eosin Stain. Magnification 25x.

usually large (2 to > 40 cm (mean 17 cm) tumours in the retroperitoneum (2 cases), paratesticular-inguinal region (3 cases), mediastinum (1 case), lung (1 case), abdomen (1 case), and popliteal fossa (1 case). The nine cases were considered to have qualified as L-LMS and showed typical WDL, with a multifocal, gradual transition into smooth muscle areas. The latter areas accounted for a variable portion of the lesion (range 5–90%) and were of low cellularity, mild to moderate atypia, and low mitotic activity. Folpe and Weiss[9] were of the opinion that these areas seemed to arise from or blend with the smooth muscle in the walls of large vessels within the tumour. One case showed areas of dedifferentiation consisting of actin and desmin-negative, high grade sarcoma. Folpe and Weiss[9] observed that follow-up in seven cases (range 26–312 months; mean 119 months) showed multiple local recurrences in seven patients and no metastases. Three patients at the time of reporting were without evidence of disease (follow-up duration 26–312 months; mean 144 months) and four patients were alive with progressive disease (follow-up duration 60–132 months; mean 99 months). Folpe and Weiss[9] concluded that their study suggested that:

- The LMS component, which is typically low grade, does not adversely affect the overall behaviour of the tumour, which is similar to that of conventional WDL.
- L-LMS should not be misconstrued as evidence of low-grade dedifferentiation, a phenomenon that identifies a more unstable and potentially metastasizing lesion.

It has been stated that the presence of abundant inflammation is uncommon in well differentiated liposarcomas (WDLPS).[10] However, Kraus and associates reported well differentiated inflammatory liposarcoma at many sites including paratesticular region.[10]

Our case did not have such inflammation. Kraus and associates also reported that the median age (62 years) and tumour location (six retroperitoneal, three head/neck, one paratesticular) was comparable to usual WDLPS, as was the fact that six of ten cases recurred locally, but none metastasized during the period of follow-up (range 13 to 228 months, median 72 months). Kraus and associates[10] summarised the key histological features of WDLPS as: (1) nodular lymphoplasmacytic aggregates; (2) intervening paucicellular stroma containing fibroblastic elements, frequently with plasma cell-rich zones and scattered atypical, often multinucleate cells, (3) merging of atypical adipocytic and inflammatory elements; and (4) adjacent clearly defined zones of lipoma-like or, more rarely, sclerosing -type liposarcoma, Kraus and associates[10] also observed that in recurrences two cases "reverted" to purely lipoma-like liposarcoma, and two cases dedifferentiated. Immunoperoxidase studies of the tumours examined by Kraus and associates demonstrated that B cells predominated in the lymphoid aggregates, and that the abundant plasma cells were were polytypic in nature. Kraus and associates supported the use of the term well-differentiated inflammatory liposarcoma for tumours of this type,

which they felt should be distinguished from high-grade liposarcoma containing inflammatory cells, and in order to identify a subgroup of WDLPS at high risk of misdiagnosis.

Dedifferentiated liposarcoma is uncommon, and only a small number of cases have been documented. McCormick and colleagues[11] described the clinicopathologic features in a series of 32 cases. All the patients were adults with an age range of 37 to 83 years (median, 67 years); 20 were men and 12 women. Fifteen cases arose from the retroperitoneum (the commonest site); six cases arose in the limbs; four in the paratesticular region; three in the peritoneal cavity; two on the trunk and one each in the buttock and larynx. One primary tumour was subcutaneous. Thirty tumours arose de novo (i.e., combined with well-differentiated tumour in the primary lesion), while 2 developed in recurrences of a previously well-differentiated liposarcoma. It was observed that the well-differentiated component was most often lipoma-like and typically there was a histologically abrupt transition to spindle celled non lipogenic tumour. The dedifferentiated component was most often observed to resemble either storiform "malignant fibrous histiocytoma" ("MFH") with limited pleomorphism or myxofibrosarcoma (myxoid "MFH"); the latter pattern is rarely otherwise seen in the retroperitoneum. A small number of the tumours exhibited appearances reminiscent of myxoid embryonal rhabdomyosarcoma. Three cases had an unusual feature in that there was the occurrence of a discontinuous micronodular pattern of dedifferentiation. After an average follow-up of 5.6 years (range, 3 months to 33 years) in 27 cases McCormick and colleagues[11] observed local recurrence in 14 patients and systemic metastases in only four patients. The primary sites of the metastasising cases were upper back, thigh, retroperitoneum and paratesticular region. There were only seven tumour-related deaths. Good prognosis in the de novo dedifferentiated liposarcoma appeared to be unrelated to the extent or morphologic pattern of dedifferentiation. Nevertheless, high mitotic activity in the dedifferentiated component was observed to be associated with a more aggressive clinical course. McCormick and colleagues concluded that[11]:

- Their study underlines that dedifferentiation in peripherally located or even subcutaneous liposarcomas does occur, albeit rarely, and that dedifferentiated liposarcomas of the limbs may metastasize.
- The results suggest that dedifferentiated liposarcomas, as a subgroup among the "MFH-like" sarcomas, have a better prognosis than pleomorphic sarcomas as a whole.

Most patients with liposarcoma of the spermatic cord present with a scrotal mass, which is usually not associated with any inguinal complaints and increases in size rather slowly over a period of months or years. Liposarcoma is a disease of the older age group.[12]

So far, no specific diagnostic procedures for the evaluation liposarcoma of the spermatic cord, has been recommended.[13] Ultrasonography provides little information on paratesticular sarcomas, in view of the fact that some are visualised as homogenous and isoechogenic, others as in homogenous and echodensity is quite variable[13]. It has been stated that the use of CT scans is not widely reported but appears to be promising, as liposarcomas are of low density and can be well demarcated but no pathognomonic features for differentiation of benign versus malignant masses are defined.[14] It has also been stated that the use of MRI scan provides good information on the local situation, but an exact evaluation of any masses cannot be obtained.[13]

Of increasing malignancy the World Health Organisation (WHO) has classified liposarcomas into the following histological groups:

- Well-differentiated liposarcoma;
- Myxoid liposarcoma;
- Round cell (poorly differentiated myxoid) liposarcomas;
- Pleomorphic liposarcoma;
- Dedifferentiated liposarcoma.

The WHO classification divides well-differentiated liposarcoma into three subtypes, lipoma-like, sclerosing, and inflammatory, without defining how much fibrous tissue is required, to place the tumour in the sclerosing category rather than the lipoma-like category or how much inflammation is required for inflammatory type.[15]

Three grades are distinguished by the American Joint Committee of Cancer: G1 = well differentiated forms; G2 = moderately differentiated forms; G3 = poorly differentiated forms. With regard to grading, well-differentiated tumours, of poor cellularity, poor vascularisation, abundant stromal connective tissue, minimal necrosis and less than 5 mitoses per field are considered of low grade.[16] Poorly differentiated tumours of rich cellularity, hypervascularization, minimal stromal connective tissue, diffuse necrosis and more than 5 mitoses per field are classified as high grade.

Microscopic examination of liposarcomas usually reveal atypical cells with large, hyperchromatic nuclei, within fibrous septa or fat, marked variation in adipocytes size; usually lipoblasts.[8]

More than one grading system is currently employed in the grading of sarcomas. The most important ones are the American by Costa and associates[17], and the European by Trojani and associates.[18] Both distinguish three malignant grades. The former is founded on the histotype and the degree of tumour necrosis, whilst the latter is based upon the degree of necrosis, the degree of morphological differentiation and the mitotic index. It is recommended that the pathologist must always provide the clinician with the indication of the malignancy grade. Nonetheless, there are some histiotypes which are "automatically" grade 1: myxoid or well differentiated liposarcoma, dermatofibrosarcoma protruberans. Myxoid fibrous histiocytoma is usually a grade 2 sarcoma. Other histiotypes including pleiomorphic liposarcoma are "automatically" grade 3.

The differential diagnosis of paratesticular liposarcomas includes the following:

- Extension from primary retroperitoneal sarcoma,
- Well differentiated tumours resemble benign fatty tumours (no atypical cells)

- Inflammatory liposarcoma resembles lymphoma (monoclonal, usually B and T cells and inflammatory fibrous pseudotumour (spindle cells usually bland),
- Sclerosing liposarcomas resemble fibromatosis (more cellular, no atypia, denser collagen, CD34 negative).[8]
- Inguino-scrotal hernia.

The general guidelines of sarcoma require that a radical surgical excision of the tumour should be undertaken. Inguinal radical orchiectomy is the suggested standard approach for sarcomas of the spermatic cord in general with wide resection margins.[19]

Some authors are in favour of a hemiscrotectomy in addition to the inguinal orchidectomy.[20] Catton advised that a second resection should be performed if the margins are positive due to the fact that frozen section is not entirely reliable.[21] Local radical excision alone appears to be insufficient for liposarcomas due to the fact that local recurrence is a major problem which occurs in 50% of the patients.[20, 22, 23]

It has been established for liposarcomas of the extremities, that the histological sarcoma type, the level of differentiation, as well as the size of the tumour have little influence on recurrence rate. On the contrary, the resection status has an impact on local recurrence.[24] Some authors have recommended adjuvant radiation due to the fact that a negative resection status can rarely be ensured.[22,23,25] Schwartz and associates have stated that in view of the radiosensitivity of liposarcomas the approach of radiation treatment is feasible.[26]

Catton and associates[21] have suggested that in view of the relative resistance against chemotherapy, there is no justification for a routine adjuvant systemic therapy in liposarcomas of the spermatic cord or any other spermatic cord sarcomas. Schwartz and colleagues[26] have stated that in contrast to other sarcomas of the spermatic cord, there has been no report of metastatic disease of liposarcomas of the spermatic cord.

The important lesson learnt from this case, is the fact that a moderate sized lump that was considered to be a benign lipoma of the cord after eight years turned out eventually to be diagnosed as Paratesticular liposarcoma. The question that may be asked is "should all clinically diagnosed lipomas be excised in order to avoid missing a liposarcoma?" Perhaps it could be said that all large clinically diagnosed lipomas should be excised if the patient's general condition permits it. A number of small lipomas may be left alone if they are not causing any problem to the patient. Nevertheless, if there is a sudden increase in size of the clinically diagnosed lipoma or in order words if the lipoma is growing faster in size than previously observed the lipoma must be completely excised.

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