

# SPINDLE CELL LIPOMA OF THE SPERMATIC CORD - A REPORT WITH REVIEW OF LITERATURE

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## ABSTRACT

A case of spindle cell lipoma of the spermatic cord is reported in a 60-year old Saudi male who presented with a one-year history of painless, firm, right testicular mass provisionally diagnosed as a testicular tumour. The final diagnosis of spindle cell lipoma of the cord was made following histopathology of the excised tumour. A high degree of suspicion and attention to clinico-pathologic features are necessary for accurate diagnosis and avoidance of unnecessary radical surgery. This may be aided by immuno-histochemical tests for vimentin, a tissue-specific, intermediate filament protein (IFP), on the excised tissue. A brief review of the literature is undertaken. To the best of our knowledge, this is the first case of spindle cell lipoma of the spermatic cord reported.

**KEY WORDS:** Spindle-cell lipoma; spermatic cord; testicular mass.

## CASE REPORT

A 60 year-old Saudi male, presented with a one-year history of a slowly growing, painless right scrotal mass. He was well otherwise, with no history of trauma, sexually transmitted infection, weight loss or sudden change in the size of the mass. There was also no associated past history of tuberculosis.

Clinical examination showed a well-looking late middle aged gentleman with normal systems. Both testicles were fully descended and intra-scrotal in location. The right testicle was associated with a non-tender, firm mass measuring 10cm x 7cm x 5cm, with a smooth surface. The mass was inseparably attached to the right testis and epididymis. There was no associated inguinal hernia. Digital rectal examination (DRE) revealed a firm benign-feeling prostate gland estimated to weigh about 30gms.

Investigations such as routine blood chemistry, alpha-fetoprotein (AFP), beta-human chorionic gonadotropin (b-hCG), Chest X-ray, and erythrocyte sedimentation rate (ESR) were normal. There was also no intra abdominal abnormality on ultrasound (U/S) and computed tomography (CT) examinations of the abdomen. Ultrasound of the scrotum revealed a large, well-defined homogeneous uniformly echogenic mass in the right hemi-scrotum. The right testis was displaced caudally and was not infiltrated by the mass, suggesting a benign lesion. The right epididymis also appeared normal. The left testis and epididymis were normal.

A provisional diagnosis of right para-testicular tumour was made. Though suspected to be a benign lesion from the clinical features, exploration and frozen section histology of the tumour was planned through an inguinal approach before definitive surgery at the same sitting. But the patient rejected this procedure, insisting instead on orchidectomy regardless of the final histology report. A radical orchidectomy was therefore performed through a right inguinal approach. The right testis was found to be adherent to the tumour but there was no gross

evidence of tumour invasion. There was no associated hydrocele. Histo-pathological report of the tumour revealed that it was well circumscribed, with an intricate mixture of mature adipocytes and interspersed with spindle cells within a mucinous matrix. There were strands of collagen fibres within the tumour substance, with no features of malignancy. A diagnosis of spindle cell lipoma of the spermatic cord was made based on these features. The resected margin of the spermatic cord was also free of tumour. More than five years following resection, there is no sign of tumour recurrence locally or of any metastatic lesion associated with the tumour.

A thorough Medline search of the literature was carried out from 1984 to 2002 but we could not find any previous description of this lesion involving the spermatic cord. We therefore assume that this is the first report in the literature, of spindle cell lipoma specifically affecting the spermatic cord. This adds one more variant to this uncommon condition, hence the report hereby presented.

## DISCUSSION

Most solid testicular masses are usually regarded as malignant until proven otherwise, while most tumours of para-testicular origin are considered benign. Sonographically, although most echogenic masses are usually benign, the potential for malignancy cannot always be totally excluded. Frozen section histology is therefore usually employed to clear any lingering doubts in such circumstances. This was the planned procedure before our patient insisted on having his testicle removed, presumably because of lingering fears of malignancy, and would not be persuaded otherwise. Spindle cell lipoma is a specific type of lipoma which can easily be confused with a liposarcoma (Enzinger and Harvey, 1975). Unlike liposarcoma however, it is a distinctly benign lesion. Since its first classic description by Enzinger and Harvey in 1975, several variants have been described including a pseudoangiomatous variant

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(Hawley, et al., 1994), a fibrous spindle cell lipoma (Diaz-Cascajo et al., 2001), a vascular variant in an intramuscular (subfascial) location (Lidang et al., 1990), and another rare variant characterized as fibrohistiocytic lipoma (Marshall-Taylor et al., 2000). Pleomorphic lipoma is also accepted as part of the spectrum of spindle cell lipoma (Fanburg-Smith et al., 1998). Multiple lesions though rare, have also been reported, varying between 2 and 220 lesions in the same patient; familial and non-familial cases have been reported (Fanburg-Smith et al., 1998).

Spindle cell lipoma of the cord is rare. A Medline search of the literature from 1984 to 2002 failed to yield any report of spindle cell lipoma affecting the spermatic cord. We however found a single report of Leydig cell tumour of the testis with adipose differentiation containing spindle cells (Ulbricht et al., 2002). This is however, a different entity from spindle cell lipoma and should not be confused with the case being reported.

More than 75% of spindle cell lipomas occur in males aged 45 to 70 years and in most cases, affect the regions of the shoulder and the back of the neck almost exclusively (Enzinger et al., 1975; Fletcher et al., 1987). It accounts for about 1.5% of adipocytic neoplasms and is outnumbered by conventional benign lipomas by approximately 60 to 1 (Fletcher et al., 1987). Lesions have been described at various sites including the limbs, face, trunk, skin, skeletal muscle, orbit and vascular tissue. Two main types of the lesion involving dermal tissues have also been described, namely, one with a plexiform pattern and the other with a nodular pattern. The plexiform pattern is predominantly seen in the thigh-groin-gluteal area and the nodular types in the head-neck or acral region (Zelger et al., 1995). The case being reported did not conform to this variant of spindle cell lipoma.

The first classic clinico-pathologic description of this tumour was that of an intricate mixture of mature adipocytes and uniform spindle cells within a matrix of mucinous material traversed by a varying number of birefringent collagen fibres (Enzinger and Harvey, 1975). Later, immuno-histochemical studies suggested that the filaments in the cells of this tumour contain vimentin (Ito and Tsuda, 1985), characterized as an intermediate filament protein (IFP) with molecular weight 52,000, found in most differentiating cells and certain fully differentiated ones. It is tissue-specific, developmentally regulated, and probably serves a structural function in the cytoplasm (Vimentin, 1996). It is also believed to play a role in communication and transport between the cell surface and the nucleus, by interconnecting the nucleus with the plasma membrane (Vimentin, 1996).

The finding of vimentin in spindle cell filaments has been subsequently confirmed by other workers who have routinely shown the tumour cells to be vimentin-positive (Marshall-Taylor et al., 2000; Zelger et al., 1995; Mehregan et al., 1995). Vimentin-positivity is currently a criterion for diagnosing spindle cell lipoma. Although we did not have electron microscopic or immuno-histochemical facilities for specific characterization of vimentin at the time of diagnosis, the tumour in our patient had the light microscopic histo-pathologic

features which qualified it as spindle cell lipoma, in keeping with the classic description of Enzinger & Harvey, who described a tumour containing "an intricate mixture of mature lipocytes and a uniform arrangement of spindle cells within a matrix of mucinous material, traversed by a varying number of birefringent collagen fibres" (Enzinger and Harvey, 1975).

The exact nature and pathogenesis of this tumour is not yet fully elucidated. It has been suggested that the accumulated filaments and myxoid matrix containing hyaluronic acid in spindle cell lipoma may be degenerative rather than synthetic in nature (Ito and Tsuda, 1985). It is now known however, that vimentin is a tissue-specific, developmentally regulated member of the intermediate filament protein (IFP) family found in most differentiating cells, particularly of mesenchymal origin, including those grown in tissue culture (Vimentin, 1996). It is expressed in early stages of cellular differentiation.

Extra-cellular vimentin has been described, which is secreted by macrophages in response to pro-inflammatory signaling pathways and is probably involved in immune function (Nirit Mor-Vaknin et al., 2002). It is also involved in bacterial killing and the generation of oxidative metabolites, two important functions of activated macrophages (Nirit Mor-Vaknin et al., 2002). Its secretion is known to be blocked by the anti-inflammatory cytokine interleukin-10 (IL-10) (which inhibits protein kinase-C [PKC] activity), and is triggered by the pro-inflammatory cytokine tumour necrosis factor-alpha (TNF- $\alpha$ ). These findings provide probable evidence that the pathogenesis of spindle cell lipoma has inflammatory and immune basis. Unlike liposarcoma, spindle cell lipoma is distinctly a benign lesion, completely amenable to local excision without recourse to radical surgery.

The planned frozen section histology for our patient should have obviated the need for radical surgery. But this must be underscored by the difficulty that exists in distinguishing this tumour from liposarcoma without the benefit of vimentin reactivity, particularly using a frozen section specimen. The patient's choice of radical surgery in this case, though surprising, was however the overriding reason in favour of radical excision. His decision must have been influenced by his understanding that certain tumours in the testicle could be malignant. There have been no adverse sequelae in the patient five years after its resection.

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