

## Primary Osteosarcoma of the Spermatic Cord: Case Report and Literature Review

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**Abstract.** Primary osteosarcoma of the spermatic cord is a rare tumour with few mentions in the literature. A 59-year-old man presented with a large painless left inguinal and scrotal mass. The patient underwent excision of the mass, which arose from the spermatic cord. A left high dissection of the spermatic cord and radical orchiectomy due to associated atrophy of the left testicle were performed. Pathological findings were suggestive of spermatic cord osteosarcoma. The patient died eleven years later of metastatic lung disease. Spermatic cord osteosarcoma is an uncommon neoplasm and its preoperative diagnosis is very difficult. Any palpable suspicious mass of the cord should be investigated with ultrasonography before excision; CT scan and magnetic resonance imaging may be helpful in defining preoperative diagnosis and the extension of the mass into the neighbouring tissues. Surgical treatment of spermatic cord sarcomas in adults is via a radical orchiectomy with high dissection of the spermatic cord and en bloc excision of involved neighbouring tissues; overall 5- and 10-year survival rates are reported in the literature to be 75% and 55%, respectively.

Primary osteosarcoma of the spermatic cord is a rare tumour with few mentions in the literature (1-3). This neoplasm is part of the wider group of paratesticular sarcomas which arise from cord tissues and must be distinguished from primitive testicular tumours. Only a

small series of spermatic cord sarcomas from single institutions have been published thus preventing the development of a consensus concerning optimal surgical strategy and possible adjuvant treatment (4-6). This paper reports a case of primary osteosarcoma of the spermatic cord; clinical and pathological features are discussed with a review of the literature.

### Case Report

A 59-year-old man was admitted in April 1989 with a five-year history of a painless bilateral groin lump. The medical history was unremarkable. Physical examination revealed a large non-tender hard mass, which involved the left groin and the scrotum. In the lower extremity of the mass there was a soft nodular structure, suggestive of an atrophic testis. A concomitant right inguinal hernia was also present. Preoperative ultrasonography (US) and a computed tomography (CT) scan showed a solid, non-homogeneous calcified oval mass which measured 15 cm in diameter and was attached to the spermatic cord, with distal displacement of the testis. A left inguinal access was performed and the mass was found to arise from the spermatic cord.

The patient underwent radical left orchiectomy, cremaster and external oblique fascia excision and high dissection of the spermatic cord. Due to strength adhesions between the mass and the scrotum itself a left hemiscrotectomy was also performed. A right inguinal hernia repair was carried out through a separate inguinal access. Recovery was uneventful and the patient was discharged on the third postoperative day. Gross pathological examination showed a capsulated mass (length 25 cm, diameter 12 cm) arising from the spermatic cord and in continuity with the testis. The mass had a hard consistency and the cut surface showed multiple focal hemorrhagic and necrotic areas within a bone-like tissue. Microscopic examination disclosed a malignant osteogenic

**Abbreviations:** CT, computed tomography; US, ultrasonography; MRI, magnetic resonance imaging.

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tumour; malignant cells were predominantly spindle, exhibiting nuclear pleomorphism and hyperchromasia, some giant multinucleated cells were also seen. The mitotic index was elevated. The extracellular matrix was predominantly composed of trabeculae of immature bone and definite osteoid foci; some trabeculae showed pronounced mineralization. Foci of chondroid matrix were also present. Multiple foci of necrosis were seen. The histopathological features were in keeping with conventional osteosarcoma with negative surgical margins (Figure 1).

The patient received postoperative external radiotherapy to the inguinal and pelvic field with a regimen of 60 Gray (Gy) for 6 weeks. The patient was followed up regularly each 6 months and he remained disease-free for nine years. At that time he developed a pulmonary metastasis and was treated with a superior left pulmonary lobectomy; nevertheless he died two years later from massive lung metastatic disease.

## Discussion

Extraskeletal osteosarcomas are rare tumours that occur more frequently in the lower (46.6%) than the upper (20.5%) extremity and in the retroperitoneum (17%) (7). Adult spermatic cord sarcomas account for approximately 30% of all genitourinary sarcoma, and they derive from mesodermal tissues of the cord. In a recent series, the most common tumour types reported were liposarcoma (51%), leiomyosarcoma (19%), embryonal rhabdomyosarcoma (13%) and malignant fibrous histiocytoma (11%) (4). Spermatic cord osteosarcoma is an uncommon tumour with only three cases mentioned in the literature (1-3). In one of these previous three cases, the tumour was a malignant mesenchymoma presenting with different pattern areas of malignant osteoid, chondromatous and liposarcomatous differentiation (3). The second reported case was a synchronous one with bilateral renal cell carcinoma and sporadic neurofibromatosis type I (2). A history of previous trauma or radiation therapy at the tumour site has been reported to increase the risk of extraskeletal osteosarcoma. In the present case, the patient did not present such risk factors (7).

Preoperative differential diagnosis appears to be difficult and involves more common intrascrotal processes such as inguinal hernia, cord lipoma and testicular masses. Any palpable suspicious mass of the cord should be investigated with US before excision; CT scan and magnetic resonance imaging (MRI) may be helpful in defining preoperative diagnosis and the extent of the mass in the neighbouring tissues (8). In the present case, MRI was not performed since the patient was treated more than 15 years ago in 1989 and at that time MRI was not available at our institution. An inguinal approach is always recommended, intraoperative

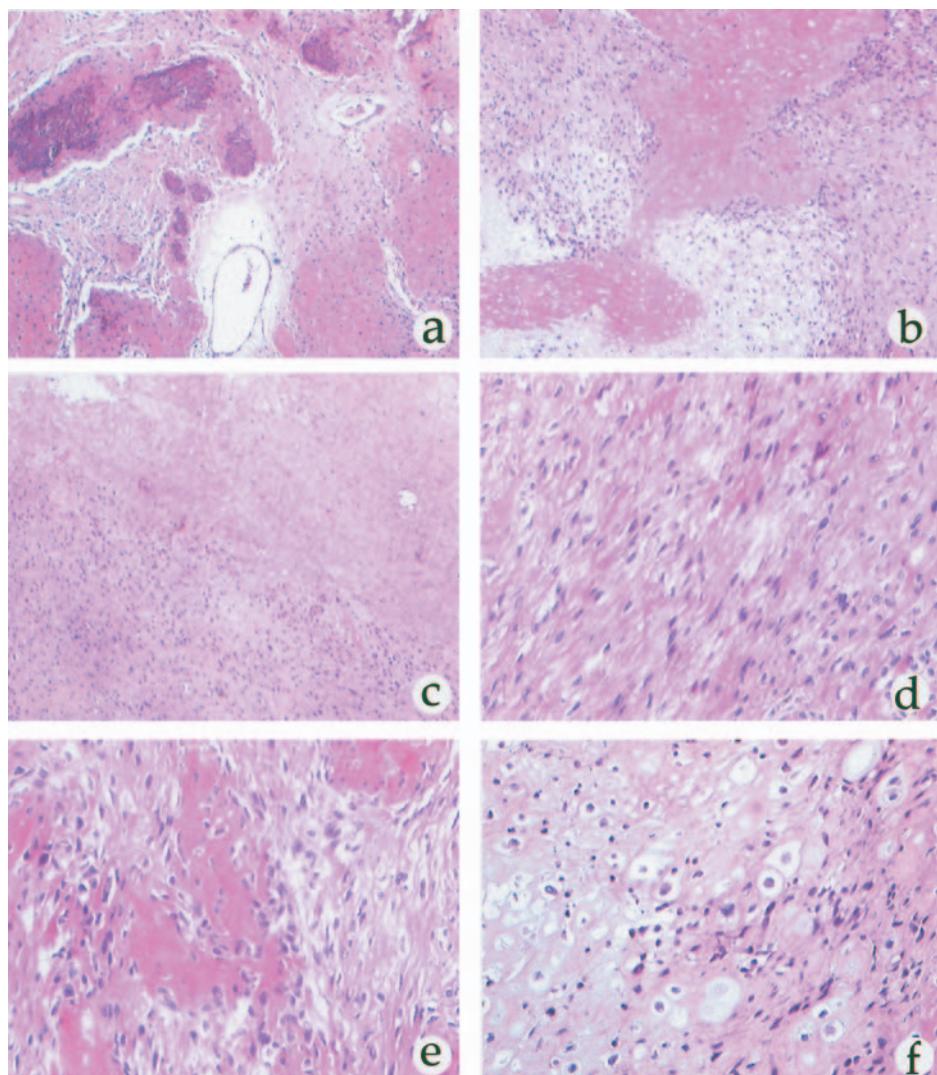
frozen sections of the mass may be of some help in surgical decision-making (1, 4). In the reported case, a frozen section was not performed since it was a bulky tumour which was impossible to separate from the spermatic cord; an associated testicular atrophy was also present.

Complete excision of the tumour with radical inguinal orchietomy and high ligation of the cord was the main primary surgical procedure (1, 2, 6). Radical orchietomy and local excision were the performed procedures in a series of 18 cases of spermatic cord sarcoma (10). All structures of the cord, including possible involved adjacent soft tissues must be removed. An optimal local control is reported to be better for T 1 lesions (<5 cm in diameter) (6, 9). In the present case excision was limited to apparently involved tissues such as the scrotum, cremaster muscle and oblique external fascia, without extending to posterior soft tissues. A more aggressive surgical policy in the management of spermatic cord sarcomas has recently been proposed as primary operation, involving wide excision of surrounding soft tissues and re-excision in case of local recurrence of the disease, since of the patients without clinically apparent disease who underwent re-excision, residual tumour was discovered in nearly one third of cases (4). Hemiscrotectomy is performed in the case of involvement of the scrotum or in patients presenting with a scrotal scar (4).

Lymphatic spread to regional nodes (ipsilateral external iliac, hypogastric, common iliac and paraaortic) has been reported in cases of synovial cell, rhabdomyosarcoma and epithelioid sarcoma (10, 11). Prophylactic retroperitoneal lymph node dissection is not routinely performed but it is mandatory for rhabdomyosarcoma or in those patients with preoperative evidence of retroperitoneal lymph node metastasis (4).

The role of adjuvant radiation therapy and chemotherapy in the management of spermatic cord sarcomas remains controversial (1, 4, 6). An improvement in locoregional control and disease-free survival has been observed following adjuvant radiotherapy. Five-year disease-free survival rates were 56% and 100% in case of surgery alone or followed by adjuvant radiotherapy, respectively ( $p < 0.01$ ) (10, 12). A multimodality approach including surgery and radiation therapy might be the most suitable strategy in order to reduce the local recurrence rate; unfortunately, there is no series large enough to support this approach and prospective trials are precluded by the rarity of this tumour (6). A doxorubicin-based chemotherapy has been used for rhabdomyosarcoma or selected cases of unresectable disease (4, 13).

In the present case, the patient received postoperative external radiotherapy in order to reduce the risk of local recurrence of the disease; he died 11 years later from metastatic lung disease, thus supporting our therapeutic



**Figure 1.** Low-power photomicrographs show a tumour characterized by production of osteoid matrix which is partly mineralized (a); a chondroblastic component (b) and areas of necrosis are present (c). High-power photomicrographs show the spindle cell component (d), osteoid deposition (e) and the chondroblastic component (f).

strategy, even if a single case result may not be enough to fully support our conclusions. Surgical treatment of adult spermatic cord sarcomas can provide an overall 5- and 10-year survival rate of 75% and 55%, respectively; an overall survival rate of 52% at 15 years has also been reported (4, 6). Due to the rarity of spermatic cord osteosarcoma, specific survival rates are not available.

### Conclusion

Spermatic cord osteosarcoma is a rare tumour, but should be considered in the differential diagnosis of inguinal and scrotal masses. Any suspicious palpable mass of the

spermatic cord should be investigated with preoperative US and MRI. Intraoperative frozen sections may be helpful to define diagnosis and to perform surgical treatment which consists of a radical orchiectomy, high ligation of the spermatic cord and complete excision of involved surrounding tissues. Inguinal orchiectomy and high ligation of the spermatic cord alone seems to be insufficient surgical treatment these days (4). Retroperitoneal lymph node dissection is reserved for preoperative evidence of lymph node metastasis and diagnosis of rhabdomyosarcoma (4). Radiation therapy may decrease local recurrence rates, in contrast the role of adjuvant chemotherapy remains uncertain (4, 10).

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