

## Case Report

# Primary Testicular Osteosarcoma – A rare case

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### Summary

A 73-year-old previously healthy man presented with a history of right sided painless scrotal lump of two-month duration. As there was clinical and radiological suspicion of malignancy he underwent right sided orchiectomy. Macroscopy revealed a solid tan testicular mass with another similar mass proximal to the resection margin of the spermatic cord. Both masses revealed similar histology of a high-grade sarcoma favouring a primary osteosarcoma. Primary testicular sarcomas are rare of which intra testicular osteosarcomas are extremely rare as reported here in.

**Key words:** *extraosseous, testicular, osteosarcoma*

### Introduction

Sarcomas of the genitourinary tract are uncommon in adults representing less than 2.7% of all sarcomas [1]. Primary intra testicular osteosarcomas are extremely rare. To our knowledge, the tumour in our patient represents the fifth reported case of an intra testicular osteosarcoma [2]. Due to its rarity the histogenesis, treatment and survival rates are not well established.

### Case report

#### Clinical history

A 73-year-old previously healthy man presented with a history of a painless scrotal lump of two-month duration. There was no history of past urogenital infection, trauma or

surgery. His past medical and surgical history was unremarkable and there was no significant family history. He was a driver by profession and neither smoked cigarettes nor consumed alcohol.

On examination, he was not pale or emaciated. There were no skeletal masses. A few enlarged right sided inguinal lymph nodes were detected. System examination was unremarkable. Scrotal examination revealed a right testicular non-illuminating hard mass measuring 3x2x2 cm in size. The left testis was normal.

His full blood count, liver and renal function tests and chest x-ray were normal. Abdominal ultrasound scan was unremarkable. Ultrasonography of the right testis showed a solid heterogenous mass measuring 3.5x3x4 cm suspicious for malignancy. As there was clinical and radiological suspicion of malignancy a radical inguinal orchiectomy was performed. The post operative period was uneventful. The patient was lost to follow up.

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Figure 1: The gross appearance of the resected specimen showing solid tan to white tumour in the testis (small arrow) and the tumour mass in the spermatic cord (long arrow).

### Pathological features

Macroscopically the specimen comprised a testis (4x2.2x2.5cm), epididymis (4x1.8x1cm) and the spermatic cord (8cm in length). Cut sections of the testis showed a solid tan to white tumour (3.5x2.5x2cm) with spotty necrosis. The tunica vaginalis and albuginea were smooth. The spermatic cord showed a similar mass (3x2.4x2cm) located 1cm proximal to the resection end (Figure 1).

Microscopy of the testicular tumour revealed a neoplasm composed of spindle cells arranged as diffuse sheets in a myxoid stroma with fibrous septae (figure 2A). The neoplastic cells had hyperchromatic, pleomorphic nuclei and scanty cytoplasm (Figure 2B). Focal areas of malignant osteoid was identified on extensive sampling

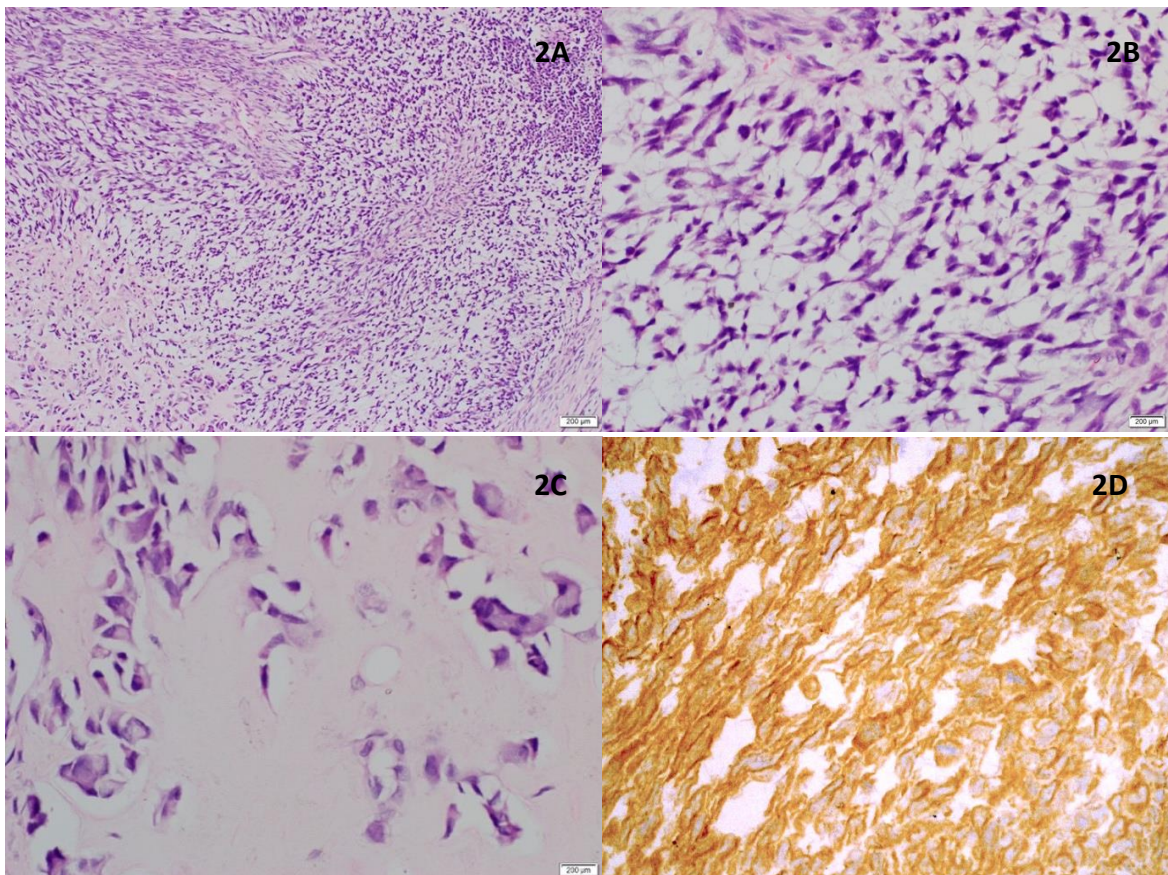


Figure 2: Microscopic findings 2A: neoplasm composed of spindle cells arranged as diffuse sheets in a myxoid stroma with fibrous septae (H & E x10). 2B: neoplastic cells with hyperchromatic, pleomorphic nuclei and scanty cytoplasm (H & E x 40). 2C: malignant osteoid (H & E x 40). 2D: Tumour cells positive for vimentin immunohistochemistry (x 40)

(Figure 2C). Mitotic activity and foci of necrosis were noted. Infiltration of the epididymis was evident. The spermatic cord mass showed similar morphology suggestive of a deposit from the testicular tumour. Elements of germ cell tumour or sex cord stromal tumour was not seen. The resection end of the spermatic cord and the tunica albuginea were free of tumour. Markedly atrophic testicular tissue was identified focally.

Immunohistochemistry revealed a strong cytoplasmic positivity for vimentin in tumour cells (Figure 2D). Pan CK, EMA, SMA, Desmin, Bcl2, CD99, CD117, CD30, Alfa feto protein, NSE, NF1, S100 and PGP9.5 were negative. A diagnosis of a primary pure testicular osteosarcoma with deposits in the spermatic cord was rendered.

### Discussion

The microscopic appearance was that of a high-grade spindle cell malignancy of the testis. The main differential diagnoses were a malignant neural tumour, leiomyosarcoma, synovial sarcoma, a germ cell tumour with sarcomatous transformation and malignant sex cord/stromal tumour. Extensive sampling was carried out to exclude other primary germ cell tumours with sarcomatous transformation and malignant mixed sex cord/stromal tumour. Negativity of SMA and Desmin excluded a leiomyosarcoma. NSE, NF1, PGP 9.5 and S100 negativity excluded neural differentiated malignancies while Pan CK, EMA, BCL 2 and CD99 negativity excluded a synovial sarcoma. Absence of germ cell tumour components were confirmed by negativity of alfa feto protein for yolk sac tumor, CD 30 for embryonal carcinoma and CD117 for seminoma.

Presence of malignant osteoid, positivity for vimentin and negativity for smooth muscle, neural and germ cell tumour immunohistochemical markers and absence of skeletal or extraskelatal neoplasms giving rise to metastasis confirmed the diagnosis of a primary osteosarcoma of the testis.

When diagnosing extraosseous osteosarcoma, careful clinical and radiological evaluation is mandatory to exclude the possibility of a deposit from primary bone osteosarcoma. In our patient there was no previous history or current evidence of a primary bone tumour

excluding the possibility of a testicular secondary deposit.

Extra skeletal osteosarcoma is a malignant mesenchymal neoplasm that produces osteoid and is found in the soft tissue without any skeletal attachment. Compared to osteosarcoma of the bone extra skeletal osteosarcoma is rare accounting for 1-2% of all soft tissue sarcomas [3-5].

Testicular osteosarcomas have been reported in paratesticular and intratesticular locations, both of which are rare [2, 6-8]. They usually present with scrotal swelling with or without pain as was seen in this patient. In some instances, they may be misinterpreted as inguinal hernias. The four cases reported already were described in 30, 60, 63 and 78-year-old patients [2]. All tumours were found completely within the tunica vaginalis. Associated cryptorchidism or irradiation as risk factors were not reported.

The histogenesis of testicular osteosarcoma is unknown. As reported by Tazi et al, it is believed to occur as a neoplastic transformation of sequestered embryonic remnants of osteogenic tissue or primitive mesenchymal cells. Transformation of pre-existing teratomatous elements that acquire the potential for a preferential mesenchymal malignancy is another suggested mechanism [2].

Management of primary testicular sarcomas is not well established. Inguinal orchiectomy without neo adjuvant treatment was the preferred management in all reported cases. The prognosis of osteosarcoma of the testis is also unclear. However, one patient was well for 44 months following surgery for primary intra testicular osteosarcoma. [8]

Primary intratesticular osteosarcomas are very rare malignancies that should be excluded in mesenchymal testicular malignancies by extensive sampling, diligent histological evaluation and clinico-radiological correlation to exclude secondary deposits from a primary skeletal or other extraskelatal osteosarcomas. Detection and reporting of this rare entity is important as histogenesis, management, treatment modalities and prognosis are not well established.

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