

## Case Report

# Metastatic Paratesticular Leiomyosarcoma in a Young Male: A Case Report

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### ABSTRACT

Para-testicular leiomyosarcoma is rare, especially in young men. This is a case of a 16 year old male presenting with a painless progressive right hemi scrotal swelling of 1 year duration and right sided abdominal mass of 2 months duration. Radiological investigations revealed a right intra-scrotal tumor sparing the testis with abdominal metastasis. The patient had right radical orchidectomy with high ligation of spermatic cord and excision of attached scrotal skin. Histopathology revealed para-testicular leiomyosarcoma likely arising from the scrotal wall. He subsequently had eight courses of adjuvant combination chemotherapy and also radiotherapy to the right hemiscrotum. He however presented with local recurrence twelve months later and had right hemiscrotectomy and pathology revealed leiomyosarcoma. The role of chemotherapy in treatment of metastatic para-testicular leiomyosarcoma has not been well established. However, this case has shown good response to adjuvant systemic combination chemotherapy.

**Keywords:** Para-testicular, Leiomyosarcoma, Hemiscrotectomy, Chemotherapy

### INTRODUCTION

Para-testicular sarcomas describe the tumors arising within the scrotum but not of testicular origin and includes tumors arising from the epididymis, spermatic cord, tunica vaginalis and various supporting structures.<sup>1</sup> The majority of para-testicular sarcomas arise from the spermatic cord.<sup>2</sup> Leiomyosarcoma is the most commonly reported histologic type of para-testicular sarcoma with peak incidence reported to occur in the sixth and seventh decades of life.<sup>3</sup> We report a case of a young man who presented with metastatic para-testicular leiomyosarcoma, and had radical orchidectomy followed by adjuvant systemic chemotherapy and hemiscrotectomy with good response.

### CASE REPORT

A 16-year old male presented with a painless progressive right hemi scrotal swelling of 1 year duration and right sided abdominal mass of 2 months duration. No history of trauma to the scrotum, exposure to radiation or history suggestive of undescended testis. No history of weight loss. Physical examination of the abdomen revealed a mass located within the right lumbar region and encroaching the midline. It measures 14cm by 12cm. It was not tender, felt hard and slightly mobile transversely (figure 1). Examination of the external genitalia



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revealed a huge right scrotal mass, measuring 16cm by 12cm by 7cm. it was non-tender and had a mixed hard and cystic consistency. It had a point of attachment to the scrotal wall. The contralateral hemiscrotum and its contents were grossly normal.

The  $\alpha$ -fetoprotein,  $\beta$ -human chorionic gonadotropin, liver function test, renal function test and chest X-ray were all normal. Scrotal ultrasound scan revealed an oval, hypovascular, heterogenous, multiseptated intra-scrotal mass, free from the testis with mild bilateral hydrocele. Abdomino-pelvic ultrasound scan showed hypoechoic intraabdominal mass occupying the right lumbar region, and containing solid and cystic components. It measured 10.5cm by 8.6cm and is separate from the liver and the right kidney. Computerized tomography scan was not done preoperatively.

He was counselled and had right radical orchidectomy with high ligation of spermatic cord and excision of a 1cm margin of scrotal skin incorporating the ulcerated area. Intra-operative findings were those of huge right intra-scrotal mass with solid and cystic areas. The orchidectomy specimen measures 15.5 X 7 X 6cm and weigh 150g (figure 2). The point of attachment to the scrotal skin was ulcerated. The left testis appeared normal. After histopathologic examination and immunostaining, a diagnosis of para-testicular leiomyosarcoma likely arising from the scrotal wall was made (figure 3 and 4).

He was subsequently referred to radio oncologist where he had eight courses of adjuvant systemic chemotherapy (cyclophosphamide, vincristine, adriamycin and dacarbazine). After the sixth course, the intraabdominal mass disappeared clinically and radiologically (figure 4). He also had adjuvant radiotherapy of 30Gy in 10 fractions to the tumor bed, pelvic and para-aortic nodes with shielding of the contralateral testis. Post chemotherapy seminal fluid analysis before radiotherapy showed azoospermia.

The patient developed local recurrence twelve months after the primary surgery and subsequently had right hemiscrotectomy with histopathological findings of leiomyosarcoma.

## DISCUSSION

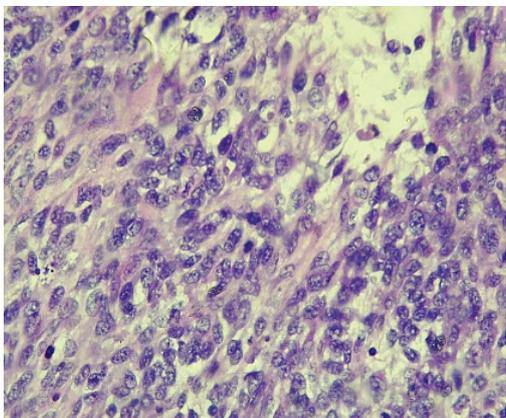
Leiomyosarcoma of the para-testicular region are rare, they have the same range of morphologic features as lesions encountered in other soft tissue sites.<sup>4</sup> Para-testicular region histogenetically consist of



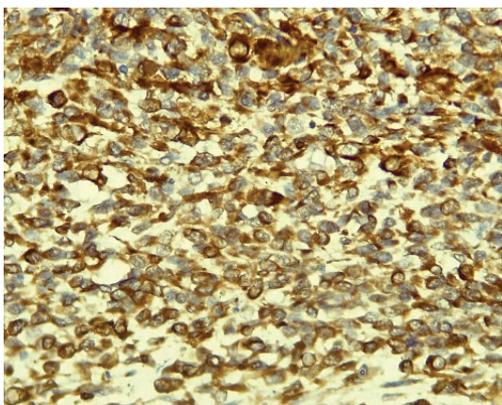
Figure 1: Pre chemotherapy image showing abdominal mass



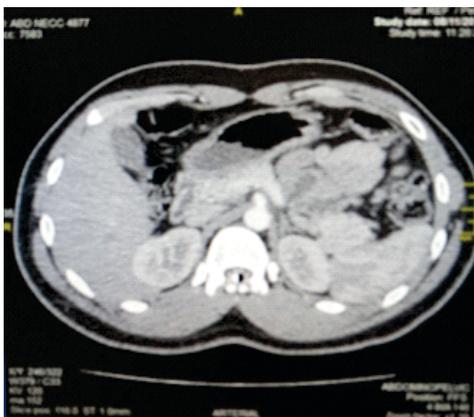
Figure 2 : Orchidectomy specimen



**Figure 3:** H & E X 100 showing spindle cells with elongated rod-shaped nuclei and abnormal mitotic figures



**Figure 4:** Photomicrograph showing positive cytoplasmic immunoreactivity to desmin



**Figure 5:** Post chemotherapy Abdominal CT scan

epithelial, mesothelial, and mesenchymal components. Thus, neoplasms developing from this region consist of heterogeneous group of tumors which demonstrate different behavioral patterns.<sup>5</sup> The para-testicular region include the testicular tunics, spermatic cord, epididymis and inguinal canal with spermatic cord being the most common site accounting for 90% of cases.<sup>6</sup> In contrast to the common site of origin, the histopathological findings in this patient showed that, the tumor is arising from the paratesticular tissue likely from the scrotal wall. Para-testicular leiomyosarcoma has a peak incidence in the sixth and seventh decade of life and usually presents as a slow-growing painless or painful discrete mass entirely separate from the testis, and sometimes associated with hydrocele.<sup>5</sup> In this case, the patient presented with a painless scrotal mass and mild bilateral hydrocele, however, presentation at a younger age was unusual when compared to the documented age at presentation. Para-testicular leiomyosarcoma is difficult to diagnose preoperatively and a definitive diagnosis requires a histologic examination of a resected specimen to observe morphological and immunohistochemical differentiation. The histology in our case revealed spindle cells with elongated rod-shaped nuclei and eosinophilic cytoplasm which was positive to desmin on immunohistochemistry.

Investigation with ultrasound scanning (USS) is the primary choice of imaging for any scrotal abnormality with a sensitivity of 95–100% for distinguishing between intra- and extra-testicular lesions.<sup>7</sup> Serum tumor markers such as alpha-fetoprotein, Beta-Human chorionic gonadotrophin and Lactate Dehydrogenase are important in the differential diagnosis of germ cell tumors.<sup>6</sup> The serum tumor markers in this case were all within normal range. All grades of sarcomas have a tendency to infiltrate local tissues and adequate initial surgical resection can be difficult, as such regional recurrence is a major problem. Scrotal recurrence rates are as high as 25–37%.<sup>8</sup> Adjuvant radiotherapy has been shown to be effective in controlling locoregional recurrence.<sup>9</sup>

Although, the role of chemotherapy in treatment of para-testicular leiomyosarcoma has not been well established, Durnali et al.<sup>10</sup> reported an 18 year old male referred to them with unresectable intraabdominal metastases 2 years after been operated for para-testicular leiomyosarcoma. Following

combination chemotherapy regimen (ifosfamide with mesna protection and doxorubicin) they achieved partial regression after 6 cycles. In our patient the intra-abdominal mass disappeared completely after the sixth course of combination chemotherapy regimen (cyclophosphamide, vincristine, adriamycin and dacarbazine) as seen from the abdominal CT scan. Complete excision of the tumor with radical inguinal orchiectomy and high ligation of the cord has been the main primary surgical procedure.<sup>11</sup> In a more recent series Ballo and colleagues reported a local recurrence rate of 30% at 10 years and 42% at 15 years after resection alone. Local recurrence being the most common pattern of failure.<sup>11</sup> Tumors in the para-testicular region tends to show local recurrence and wide resection margins can be difficult to obtain. If scrotal skin is involved, hemiscrotectomy is indicated.<sup>12</sup> Our index case presented with local recurrence twelve months after the primary surgery and he subsequently had right hemiscrotectomy. Having presented with scrotal infiltration at the time of diagnosis, our patient would have benefited from right hemiscrotectomy to reduce or prevent local recurrence. The delay in commencing adjuvant radiotherapy may have also contributed to early local recurrence in this case. Our patient is young and presented with abdominal metastasis at the time of diagnosis making it an uncommon presentation.

## CONCLUSION

In conclusion, metastatic para-testicular leiomyosarcoma in a young male is rare and treatment with adjuvant chemotherapy following radical orchidectomy is not well established. Therefore, we report our case who was treated with radical orchidectomy and adjuvant systemic combination chemotherapy with good response. In the presence of scrotal skin involvement at the time of diagnosis, hemiscrotectomy and adjuvant radiotherapy may be a better option.

**Conflict of interest:** Authors declared no conflict of interest.

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